**School of Psychology and Speech Pathology**

# Dignity Therapy: A Psychotherapeutic Intervention to Enhance the End of Life Experience for People with Motor Neurone Disease and their Family Carers

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**This thesis is presented for the Degree of**
**Doctor of Philosophy**
**of**
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## Declaration

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgement has been made.

This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

Brenda Bentley

Signature: …….

Date: ……

## Abstract

A diagnosis of motor neurone disease (MND) is a devastating experience for people with MND and their families. People with MND experience unremitting loss during the course of the disease as they gradually lose physical function and abilities. Because there is no curative treatment, care goals for people with MND and their families focus on maintaining quality of life (QOL). Research has shown that QOL for people with MND and their family carers is not related to physical decline and impairment, but rather to psychological, psychosocial and existential factors. As a result of psychosocial distress, people with MND are at an increased risk of suicide and seek out hastened death more frequently than people with other terminal illnesses.

Development of interventions that address psychological, psychosocial and existential distress in people with MND and their caregivers have often been suggested in the research literature. However, there is a lack of intervention studies and an absence of research and direction for providing this type of care. Dignity therapy, which is based on an empirical model of dignity at the end of life, was developed to reduce psychosocial and existential distress in palliative care populations and it has been shown to benefit people dying of cancer in previous research. However, these results are not transferable to people with MND as characteristics of this group differ from cancer, including baseline levels of distress, ability to communicate, physical impairment, and cognitive acuity.

This study used a cross-sectional one group pre-test post-test design to explore the feasibility, acceptability and potential benefits of dignity therapy on 29 people with MND and 18 of their family carers in Australia. For people with MND, outcomes measured hopefulness, spirituality and dignity. For MND family carers, outcomes measured caregiver burden, anxiety, depression, and hopefulness. Acceptability was measured in both groups with a self-report feedback questionnaire used in previous dignity therapy research. Feasibility was assessed by examining how common MND symptoms affected the therapy, family carers' involvement in the therapy, the length of time needed to complete dignity therapy, and any special accommodations or deviations

from the dignity therapy protocol. In addition, demographic data was collected from people with MND and their family carers, people with MND completed a health related QOL measure, family carers assessed physical impairment of the care recipient with the ALS Functional Rating Scale, and people with MND completed a cognitive screening instrument developed specifically for people with MND. Generalized linear mixed models were used to analyze the data on the group level, and reliable change scores were computed to investigate the presence of reliable pre-post change at the individual level. Descriptive statistics were used to summarize demographic variables and feedback responses. SPSS Version 20 was used.

There were no changes in the outcomes on the group level for both groups, people with MND and MND family carers. However, there were increases and decreases for both groups in hopefulness when data was analyzed on the individual level using Reliable Change scores. For MND family carers, there were also reductions in anxiety and depression for 3 family carers after dignity therapy. Dignity therapy was acceptable to both people with MND and MND family carers, who reported similar benefits on the feedback questionnaire as found in previous research with people with cancer. However, some MND family carers also expressed negative effects as a result of the intervention which require further exploration in future studies.

Dignity therapy was feasible with people with MND, though 80% of participants had symptoms that affected the intervention and required accommodation, including speech loss, speech impairment, cognitive impairment, emotional lability and paralysis. Emotional lability was encountered most often, but speech impairment presented the greatest difficulty. People with speech loss were able to complete the intervention through email, a modification to the therapy. Mild to moderate cognitive impairment had little impact on dignity therapy in all cases except one, where behavioral variant FTD was suspected. Dignity therapy took an equivalent number of sessions and therapist hours to complete with people with MND compared to figures reported in previous research, but an extended time frame was needed to complete the intervention on average.

Recommendations for the clinical use of dignity therapy with people with MND include:

- The use of screening measures for cognitive-behavioral impairment, speech impairment, and emotional lability;
- Providing choices for people with MND about how to complete dignity therapy, including the use of e-health methods; and
- Care when dealing with families who have complex histories, conflictual patterns of relating or other dysfunction, with families who are struggling to accept the terminal nature of MND, and with participants and family members from non-Western cultures.

The findings suggest that dignity therapy, an end of life intervention developed and tested for people with cancer, may also benefit people with MND and their families. However, psychotherapists need education and training about MND prior to engaging in therapy, and flexibility is needed to accommodate symptoms.

**Acknowledgements**

*Morning Dad, I read your interview on the train this morning. You have had such an amazing life! I just wanted to let you know how much it meant to me to see everything you have accomplished and overcome laid out in what I have to say is a very compelling, funny and deep interview! It almost felt like I was reading a novel, all the places you've been, the people you have met and your sometimes tumultuous spiritual journey. It was really engrossing! I wanted to get out a highlighter and start outlining great quotes – 'What is life without God? It was like looking into a black hole and I thought, No, this is not what I want for my life' – was my favourite. I won't go on and on, but that's just what I wanted to say. Love you Dad, your life is far from over, but everything you have done with the time you have had has left me quite inspired!*

There are so many people to thank for their contributions to this research.

First and most importantly, I would like to thank the study participants and their families, like the father described by his son in the excerpt above, for their willingness to be a part of this study and for their courage, commitment and dedication to seeing it through. During my research, I was welcomed into homes and at bedsides, shown to the most comfortable chair, and fed cups of tea and an occasional meal. This generosity of spirit extended into our dignity therapy work, through the biographical stories recounted, our sometimes raw and honest conversations about life's highs and lows, important lessons learned, messages to be shared, and the meaning of life. These exchanges were punctuated with the sharing of unique talents and experiences of the participants; a song written and played, a favorite Bible passage read aloud, a poem, a tour of the painting studio or a view of the beautifully tended garden. Often, I was not allowed to leave without taking home some lemons from the tree or vegetables from the garden. It was not lost on me that these acts of kindness were given by people facing the end of their lives and their families, and it was deeply meaningful to be chosen to spend time with them when their time together was limited. The generosity I encountered often continued after we finished our work together: a thank you card, news from the family when a study participant died, or the sharing of a touching note about the dignity therapy document like the one shared above. I imagine few doctoral students are given such a

rich and memorable experience through their research and I am grateful for the contributions of each and every participant and family member, the impact they have had on me, and the meaning this project has given to my life.

I have deep gratitude for my supervisors. My primary supervisor Dr. Moira O'Connor not only provided the expert guidance, insights and expertise that comes from many years' experience, but she has also been a strong advocate and consistently provided much needed support and encouragement. I could not have completed this project without her support. In addition to benefitting from her research experience, Dr. Lauren Breen has taught me about thinking ahead, thinking things through and about balance. Both Moira and Lauren made themselves available to me when needed, as much as needed, cheered me on and helped steer me in the right direction, but most importantly, they empowered me to feel that my project was important, my contribution significant, and that I could accomplish anything I set my mind to accomplish.

Bob Kane provided expert advice and assistance with the quantitative analysis. Dr. Sara Trechter read portions of the thesis and provided helpful feedback. My doctor, Alison Hogg, kept me healthy and functioning, which proved to be no small feat.

I must also thank the Motor Neurone Disease Association of WA for their support and encouragement, especially Cherylyn, Jude and Rose. Members of the Shenton Park MND clinic, including Dr. Rob Edis, Margaret Rogers and Karlene Jordan, took me under their wings and provided practical advice and knowledge about all aspects of motor neurone disease.

Thank you to my friends and family for believing in me and for being proud of my ambitions and accomplishments. My mom and Charlie, Sara, Aldine and Gali especially made me feel my work was important and that I could do it.

Thank you to my children and stepchildren. Alek and Sasha lost their Californian mother to remote Western Australia just as they entered adulthood. Kate endured a difficult international move and high school in a foreign country. Despite the cost to themselves, they were always very proud and supportive of me and they did not complain. Thomas, Sarah, Holly and John had to travel from Sydney to Perth to visit

their father because my study took us here, which was difficult, inconvenient and when they preferred us to be closer.

Finally, I would not have been able to accomplish this research without my husband, Peter. He carried most of the responsibility for a roof over our heads and food in our bellies, provided fun and rest when I needed a break, his chest to lay my head upon when I needed comforting, a constant stream of jokes to lighten the days, and his infectious, positive “can-do” attitude to keep me going. I couldn’t have done this without his patience, understanding, prayers, love and friendship. He’s been my chief cheerleader from day one. Thank you, Peter.

## List of Included Publications

Bentley B., Aoun S.M., O'Connor M., Breen L.J., Chochinov H.M. (2012) Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers? *BMC Palliative Care* 11(1):18 doi: 10.1186/1472-684X-11-18

Bentley B., O'Connor M., Kane R., Breen L.J. (2014) Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease. *PLoS One* 9(5) e96888 doi: 10.1371/journal.pone.0096888

Bentley B., O'Connor M., Breen L.J., Kane R. (2014) Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neurone disease. *BMC Palliative Care* 13:12 doi: 10.1186/1472-684X-13-12

Bentley B. (2012) It takes the time that it takes. *Journal of Palliative Medicine* 15(8), 949-950

## Statement of Contribution by Others

The purpose of this statement is to summarize and identify the nature and extent of the intellectual input by the PhD candidate Brenda Bentley and co-authors on the study publications contained herein.

Professor Samar Aoun and Professor Harvey Chochinov were involved with the study conception and securing funding for the study. They participated in the research design, and were initially involved with the project and the supervision of the PhD research before leaving the project and supervision team. They participated in writing and approving one of the publications:

**Bentley B., Aoun S.M., O'Connor M., Breen L.J., Chochinov H.M. (2012) Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers?** ***BMC Palliative Care*** **11(1):18 doi: 10.1186/1472-684X-11-18**

Dr. Moira O'Connor was co-supervisor and became primary supervisor on the PhD research replacing Professor Aoun. Dr. O'Connor was involved in the project coordination, participated in the research design, and led the supervision of the research. Dr. Lauren Breen was co-supervisor on the PhD research. Both Dr. O'Connor and Dr. Breen participated in writing and approving the three publications listed below.

**Bentley B., Aoun S.M., O'Connor M., Breen L.J., Chochinov H.M. (2012) Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers?** ***BMC Palliative Care*** **11(1):18 doi: 10.1186/1472-684X-11-18**

**Bentley B., O'Connor M., Kane R., Breen L.J. (2014) Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease.** ***PLoS ONE*** **9(5):e96888 doi: 10.1371/journal.pone.0096888;**

**Bentley B., O'Connor M., Breen L.J., Kane R. (2014) Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neurone disease.** ***BMC Palliative Care*** **13:12 doi: 10.1186/1472-684X-13-12**

Dr. Robert Kane provided assistance with the data analysis and interpretation, and participated in writing and approving the two publications listed below.

**Bentley B., O'Connor M., Kane R., Breen L.J. (2014) Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease.** ***PLoS ONE*** **9(5):e96888 doi: 10.1371/journal.pone.0096888;**

**Bentley B., O'Connor M., Breen L.J., Kane R. (2014) Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neurone disease. *BMC Palliative Care* 13:12 doi: 10.1186/1472-684X-13-12.**

I affirm the details stated in the Statement of Contribution are true and correct.

Dr. Moira O'Connor, Supervisor

Brenda Bentley, PhD Student

## List of Additional Publications and Conference Presentations

### Publications

Aoun S.M., Bentley B., Funk L., Toye C., Grande G., Stajduhar K. (2013) A 10-year literature review of family caregiving for motor neurone disease: Moving from caregiver burden studies to palliative care interventions. *Palliative Medicine* 27(5):437-46.

### Conference Presentations

Bentley, B., O'Connor, M., & Breen, L. (2013 December) *Dignity Therapy: A psychotherapeutic intervention to enhance the end of life experience for people with motor neurone disease and their family carers*. Platform presentation, International ALS/MND Symposium, Milan, Italy.

Bentley, B., O'Connor, M., Edis, R., & Vojkovic, S. (2013 December) *The end of life experience of people with MND compared to those with cancer: Family carers' perspectives*. Poster presentation, International ALS/MND Symposium, Milan, Italy.

Bentley, B., O'Connor, M., Edis, R., & Vojkovic, S. (2013 November) *The end of life experience of people with MND compared to those with cancer: Family carers' perspectives*. Poster presentation, MND Australia Annual Research Meeting, Sydney, Australia.

Bentley, B., O'Connor, M., & Breen, L. (2013 October) *Dignity Therapy: A brief psychotherapy for dying patients and their families.* "How to" session, Australian Psychological Society Annual Conference, Cairns, Australia.

Bentley, B., O'Connor, M., & Breen, L. (2013 September) *Dignity Therapy: A brief psychotherapy for dying patients and their families.* Workshop session, Australian Palliative Care Conference, Canberra, Australia.

Bentley, B., Aoun, S., O'Connor, M., Breen, L. & Chochinov H.M. (2012 October) *Dignity therapy's ability to enhance the end of life: Case examples from the dignity therapy/motor neurone disease study*. Workshop session, WA Palliative Care Conference, Perth, Australia.

Bentley, B., Aoun, S., O'Connor, M., Breen, L. & Chochinov H.M. (2012 September) *Dignity therapy's ability to effect change in patients and family: Preliminary results from the Dignity Therapy/Motor Neurone Disease study*. Australian Psychological Society Annual Conference, Perth, Australia.

Bentley, B., Aoun, S., O'Connor, M., Breen, L. & Chochinov H.M. (2012 September) *Dignity therapy's ability to enhance the end of life for people with MND and their*

*families; preliminary results from the dignity therapy/motor neurone disease study*. National MND Conference, Adelaide, Australia.

Bentley, B., Aoun, S., O'Connor, M., & Chochinov H.M. (2011 December) *Dignity Therapy for people with motor neurone disease and their family carers.* Palliative Care for Motor Neurone Disease, WA Centre for Cancer and Palliative Care 2011 Research Seminar, Perth, Australia.

Bentley, B., Aoun, S., O'Connor, M., & Chochinov H.M. (2011 November) *Dignity Therapy: A psychotherapeutic intervention to enhance the end of life experience for people with Motor Neurone Disease and their family carers: preliminary findings*. Poster presentation, International ALS/MND Symposium, Sydney, Australia.

Bentley, B., Aoun, S., O'Connor, M., & Chochinov H.M. (2011 November) *Dignity Therapy: A psychotherapeutic intervention to enhance the end of life experience for people with Motor Neurone Disease and their family carers: study protocol*, Poster Presentation, Mark Liveris Research Forum, Curtin University, Perth, Australia.

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## Figures & Tables

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## Abbreviations

AAC ..... Augmentative and alternative communication

ACAT ..... Aged care assessment team

ACT ..... Australian Capital Territory

ALS ..... Amyotrophic lateral sclerosis

ALSAQ -5 ..... ALS Assessment Questionnaire -5

ALS-CBS ..... ALS-Cognitive Behavioral Screen

ALSFRS-R ..... ALS-Functional Rating Scale

APAI ..... Australian Postgraduate Award Industry

ARC ..... Australian Research Council

BOMC ..... Blessed Orientation Memory Concentration test

FACIT-sp-12 ..... Functional Assessment of Chronic Illness Therapy -Spiritual Wellbeing Scale-12

FTD ..... Frontotemporal dementia

GLMM ..... Generalized linear mixed models

GP ..... General Practitioner

IDG ..... Inter-disciplinary group

IRCT ..... International randomized controlled trial

HADS ..... Hospital Anxiety and Depression Scale

HHI ..... Herth Hope Index

LTMV ..... Long term mechanical ventilation

MND ..... Motor neurone disease

MNDAWA ..... Motor Neurone Disease Association of Western Australia

QOL ..... Quality of life

NIV ..... Non-invasive ventilation

NCCCP.....Neurodegenerative Conditions Coordinated Care Program

PAS ..... Physician-assisted suicide

PEG .....Percutaneous endoscopic gastronomy

PDI ..... Patient Dignity Inventory

RC ..... Reliable change

RCT.....Randomized controlled trial

UK.....United Kingdom

US..... United States

WA.....Western Australia

WACCPC.....Western Australian Centre for Cancer and Palliative Care

WHO ..... World Health Organization

ZBI ..... Zarit Burden Interview

# CHAPTER ONE
# 1. Introduction

Chapter One provides an introduction to the thesis examining the feasibility, acceptability, and potential effectiveness of dignity therapy to enhance the end of life of people with motor neurone disease and their family carers. This chapter provides definitions for psychosocial care and palliative care. It presents the three main challenges facing palliative care today, which are the provision of psychosocial care, the incorporation of care and supports for family carers, and the inclusion of people with illnesses other than cancer. The researcher will be introduced and experiences of the researcher relevant to this study are shared. This chapter concludes with the study background and funding, the aims and significance of the study, and the overall structure of the thesis.

> You matter because you are you. You matter to the last moment of your life, and we will do all we can, not only to help you die peacefully, but also to live until you die. (Saunders 1976 p. 1005)
>
> Dame Cicely Saunders

## 1.1 Thesis introduction

Where can people turn when diagnosed with a life-limiting illness where they gradually become paralyzed and lose the ability to move without help, speak to friends and family, swallow their food, and breathing itself becomes difficult? How do partners and spouses cope with watching their loved ones become increasingly impaired and with becoming full-time carers? This thesis presents a study of the effects of dignity therapy, a palliative care psychosocial intervention designed to address psychosocial and existential distress, on people who have motor neurone disease (MND) and their family

carers. MND is a fatal and rapidly progressing neurodegenerative disease (to be discussed in more detail in Chapter Two). People with MND and their families experience exceptional strain from symptom onset to death and have significant unmet psychological needs. This thesis hypothesizes that dignity therapy may help to address these needs.

## 1.2 What is palliative care?

Dame Cicely Saunders, the founder of the modern hospice movement, introduced a holistic philosophy surrounding end of life care. Her influential perspective and work at St. Christopher's hospice in London emphasized that quality care at the end of life must seek to relieve all areas of pain experienced by people as they approach death when cure is no longer an option. Saunders introduced the idea of 'total pain,' which included not only physical pain but also emotional, psychosocial and spiritual dimensions of distress (Saunders 1964). Palliative care, a term originally used by Balfour Mount in the early 1970s (Mount 1997, Watson, Lucas et al. 2009), embraces the hospice philosophy. The terms *palliative care* and *palliative medicine* have become inclusive terms applied to terminally ill people and encompasses those cared for in hospice facilities, people in hospital who seek symptom relief rather than curative treatment, and people who wish to be cared for and die at home or within the community (Billings 1998). There are three spheres of concern for palliative care providers – symptom control, psychosocial care, and disease management. In order to provide holistic care, each sphere must be considered in relation to the other two, each sphere has overlap with the others, and all three spheres of concern are equally important (Watson, Lucas et al. 2009).

The World Health Organization (2014) defines palliative care as:

> …an approach that improves the quality of life of patients and their families facing the problems associated with life-threatening illness, through the prevention and relief of suffering by means of early identification and impeccable assessment and treatment of pain and other problems, physical, psychosocial and spiritual. (World Health Organization 2014 para. 1)

The US National Consensus Project for Quality Palliative Care (2014) presents this vision:

> Palliative care is both a philosophy of care and an organized, highly structured system for delivering care. The goal of palliative care is to prevent and relieve suffering and to support the best possible quality of life for patients and their families, regardless of the stage of the disease or the need for other therapies. Palliative care expands traditional disease-model medical treatments to include the goals of enhancing quality of life for patients and family members, helping with decision-making, and providing opportunities for personal growth. (National Consensus Project for Quality Palliative Care 2014 para. 1)

Palliative Care Australia (2014) provides the definition "Palliative care is specialist care provided for all people living with, and dying from a terminal condition and for whom the primary goal is quality of life" (Palliative Care Australia 2014 para. 6). What is clear from these definitions is that enhancing the quality of life of patients and family at the end of life is a paramount concern in palliative care.

The palliative care field is currently facing challenges in three major areas. The first of these challenges is providing end of life care to all people equitably regardless of disease. Modern day palliative care is deeply rooted in cancer care due to the historical influence of Dame Cicely Saunders' St. Christopher's hospice which cared for people with cancer (Watson, Lucas et al. 2009). As a consequence, the field has neglected other terminal disease populations (Potter, Hami et al. 2003) and over 90% of people in the UK currently receiving specialist palliative inpatient care have a cancer diagnosis (Watson, Lucas et al. 2009). However, palliative care practitioners recognize the need to offer care to people with other life-limiting illnesses (Rosenwax, McNamara et al. 2005) who also suffer from distressing symptoms and have extensive palliative care needs (Addington-Hall, Fakhoury et al. 1998, Murtagh, Preston et al. 2004). As such, efforts are being made to extend palliative care equitably to all people at the end of life (Kristjanson, Aoun et al. 2005, Ostgathe, Alt-Epping et al. 2011, Johnston 2014).

The second challenge is providing support to family members of the terminally ill (Aoun, Bentley et al. 2013). The holistic definition of palliative care states that the unit of care comprises the dying person and their carers and family members (Billings 1998, World Health Organization 2014). Traditionally, and in years past, the care extended to family members was bereavement support, which remains an important

component of end of life care (Watson, Lucas et al. 2009, Breen, Aoun et al. 2014). However, because the majority of people prefer to be cared for by their families and die at home (Grande, Stajduhar et al. 2009, Wilson, Cohen et al. 2013), the role of family has taken on increasing importance (Watson, Lucas et al. 2009). Home-based palliative care would be impossible for most of the terminally ill without the support of family (Hudson 2003) who relieve public institutions of substantial economic burden by providing care (Chai, Guerriere et al. 2013). Family carers often find the complex caring role stressful (Aoun, Kristjanson et al. 2005) and research has documented the significant burden placed on them (Harding and Higginson 2003, Goldstein, Atkins et al. 2006, McCabe, Firth et al. 2009). As a result, the palliative care field has increased its focus on understanding and meeting the needs of family members who provide care for the terminally ill (Grande, Stajduhar et al. 2009, Hudson and Payne 2011, Harding, List et al. 2012).

The third challenge is providing psychosocial care. Despite Dame Cicely Saunders' early definition of total pain, which includes physical, emotional, psychosocial and spiritual dimensions of distress, and ongoing calls to address total pain (Chochinov 2007), palliative care has advanced most quickly in the areas of physical pain and symptom management as these areas were easier to address using medical science and technology. Psychosocial, existential, and spiritual aspects of well-being at the end of life are less understood, but research has revealed that these types of distress are often *more* significant to a person's quality of life during a terminal illness than physical pain and physical symptom relief (Clarke, Hickey et al. 2001, Robbins, Simmons et al. 2001). Psychosocial and existential distress is related to quality of life, depression, hopelessness and suicidal ideation in people receiving palliative care (Breitbart, Rosenfeld et al. 2000, Chochinov 2006). As a result, there is an increased focus on understanding psychosocial needs at the end of life (Pulchaski 2007, Ventura, Burney et al. 2014), and more resources are being developed to provide improved psychosocial care (Breitbart, Gibson et al. 2004, Chochinov 2005, Rodin 2013, Waldron, Janke et al. 2013). Nonetheless, there are few empirically supported psychosocial interventions for use at the end of life (Hudson and Payne 2011, Pagnini 2012). These three challenges inform, and will be informed by, this thesis which details a

psychosocial intervention for people living with MND and their family carers.

### 1.3 What is psychosocial care?

Palliative psychosocial care specifically seeks to address the emotional, social, spiritual, and psychological issues that occur when a person has a terminal illness and is facing death, when emotional distress and existential suffering are commonly encountered (Breitbart, Gibson et al. 2004). Psychosocial care focuses on the psychological and emotional wellbeing of people with illness and their families to optimize quality of life: it covers a broad area including spirituality, existential concerns, self-esteem, family issues, psychological distress, social supports, relationships, and day-to-day concerns about the impacts of illness (McLeod 2003). These broad overlapping areas result in psychosocial care being vaguely defined and often poorly understood (Pulchaski 2007, Daaleman, Usher et al. 2008, Vachon, Fillion et al. 2009).

Questions such as "Why me?" "What is the purpose of life," and "What will happen after I die?" are primary existential questions for people facing death (Boston, Bruce et al. 2011). Existential psychotherapist Irvin Yalom identified death anxiety as a universal human phenomenon and theorized that facing death is not only emotionally terrifying but also a catalyst for reflection on existential meaning (Yalom 2009). Spirituality is a source of meaning for most people throughout the world and research shows it becomes even more important in people facing serious illness (Breitbart 2001, Kaut 2002, Pulchaski 2007, Vachon, Fillion et al. 2009). Dimensions of spirituality include meaning in life; connection to something greater than oneself; values, morals and beliefs; and connection to others (McGrath 2002, Pulchaski 2007). Peaceful relationships with loved ones, concerns about the impact of death on others, and relieving burden on family carers are also important psychosocial concerns at the end of life (Breitbart, Gibson et al. 2004). Significant distress in any of these areas can result in psychological distress, including depression, anxiety and suicidality (Boston, Bruce et al. 2011).

Not only is there a lack of agreement about the exact nature of psychosocial care but there is also a lack of consensus about which members of the multidisciplinary palliative care team should provide this care (Pulchaski 2007, Daaleman, Usher et al.

2008, O'Connor and Fisher 2010). Palliative care physicians and nurses are often cited as the providers of psychosocial care because they are the most closely connected to people with terminal illnesses and families (Edwards, Pang et al. 2010), but it is questionable whether clinicians can or should take on these tasks in addition to their other roles (Daaleman, Usher et al. 2008, O'Connor and Fisher 2010). Hospital chaplains and pastoral care providers are also suitable providers of psychosocial care based on their expertise in the area of spirituality and religion (Kaut 2002), but the terminally ill often underutilize these providers at the end of life (Edwards, Pang et al. 2010). Psychiatrists, psychologists, counselors and social workers also have a role to play in the provision of psychosocial care by providing counseling, psychotherapy and psychological interventions (Chochinov and Breitbart 2012), although many in these disciplines feel conflicted about the sometimes spiritual nature of psychosocial care and are reticent to approach such issues (Johnson 2003). A report from the American Psychological Association in 2000 stated, "There is little evidence that the discipline of psychology as a whole has considered end of life issues to be an important area that deserves substantial professional time and attention" (as cited in Haley, Larson et al. 2003 p. 627). Uncertainty among palliative care providers about how should provide psychosocial care may leave a large number of palliative care patients and family members with unmet psychosocial needs (Schulman-Green 2003).

### 1.4 The researcher

This section is included to share with the reader my personal research orientation and the ways my background and experiences have influenced my views and decision to research this topic. As all research has a subjective component (Ercikan and Wolff-Michael 2006) that is influenced by culture and values, it is common for researchers to clarify their personal motivation for embarking on their research (Prilleltensky 1997, Creswell 2009). This is my goal here.

My journey began in 2008 when I undertook a Master's degree in Counseling Psychology from the University of San Francisco, in California, United States (US). When our practicum began in 2009, I chose to work in a community hospice. Initially, I (along with the other counselors) performed bereavement counseling with family

members after the death of hospice patients. We held sessions in a comfortably furnished office tucked into a far corner at the hospice base, often on nights and weekends, and we rarely came into contact with the other staff and clinicians. Over time, I developed a deep curiosity about their work. Voicing that curiosity, I was invited to sit in on weekly two-hour interdisciplinary group (IDG) meetings where the case of each hospice patient was discussed. At this hospice, these meetings were attended by the hospice physician, pharmacist, nurse case managers, social workers, spiritual care worker, volunteer manager, and bereavement manager. I observed that psychosocial support was provided to the hospice patient and family primarily by the social worker, spiritual care worker, and volunteers, although everyone involved played a role to some degree. After a patient died, bereavement counselors were introduced to all significant family members, and they continued to provide grief support to family members for up to one year.

Through attending the IDG meetings, I formed an opinion that the psychosocial needs of the hospice patients and families were not being adequately met. First, I observed how everyone at the hospice was stretched in their roles. For example, there was one chaplain for 40-50 patients placed throughout the community. The chaplain's role mostly involved spiritual counseling and performing rituals such as communion. Due to practical and logistical issues services were only available to a few. Harried social workers attended to the bureaucratic needs of families such as helping to prepare advance care planning documents, assisting with caring arrangements, and planning funerals. Volunteers were called on to provide companionship, if and when they were available. As I listened to various stories in the meetings about psychosocial, existential and emotional distress experienced by both patients and carers, and heard these stories again during bereavement counseling, I wondered about my role as a counseling psychologist and the discipline of psychology more generally in helping patients and their families in the transition from life to death. I asked my supervisor if I could help, and I was given permission to counsel hospice patients and their families.

My first challenge in this new task was identifying empirically based psychotherapeutic modalities or interventions designed to alleviate distress at the end of life. The hospice framework did not involve psychologists or counselors before

bereavement, so there were very few resources. I asked several of my professors what they would do, but I received little guidance in response. One instructor memorably answered he would do nothing as the patient would soon die and instead he would concentrate on the needs of the family members. I performed a review of the literature but found very little.

The most promising intervention I encountered was dignity therapy. It was designed to be used with palliative care patients, it was brief, it showed benefit to both patients and family members, and it was based on empirical research. I began to use some of the dignity therapy questions while counseling patients, such as "Tell me about a time when you felt most alive" and "What have you learned about life?" These conversations appeared to provide therapeutic benefits as patients were engaged, uplifted, and an improved mood was observed by the end of the session. As a result, I became more intrigued about the intervention.

Around this time, my practicum was ending and graduation was near. I had to make a decision about my future plans, including whether to continue clinical work or embark on further study. On a whim, I sent an email to Professor Harvey Max Chochinov who developed dignity therapy. I shared that I was considering the pursuit of a PhD and that I was interested in dignity therapy. I asked if he knew of any opportunities in dignity therapy research. I anticipated that my email would go unanswered, but I was pleasantly surprised when Professor Chochinov offered his encouragement and responded that he knew of one opportunity in Perth, Australia. He put me in touch with the researchers, and soon I was offered the opportunity to obtain a PhD through a research project of dignity therapy with people who had motor neurone disease (MND) as it is known in Australia, or amyotrophic lateral sclerosis (ALS) as I knew it in the US.

When offered this opportunity, I reflected on my experiences counseling people with MND and their families at the hospice. These cases were some of our most challenging. One case stood out in particular. A man in his 50s who rode a motorcycle and enjoyed an active, outdoor lifestyle could not come to terms with losing his physical abilities from MND. Though he retained some mobility and could function better than

other people with MND I had encountered at the hospice, he was experiencing such psychological distress as a result of his deteriorating condition that he stopped eating and drinking to hasten his death. After a week, the palliative care physician intervened. With the agreement of the man's wife, the doctor performed palliative sedation where the man with MND was sedated to an unconscious state until he died from dehydration. I had encountered palliative sedation before but not where it was used as a solution to psychological pain rather than physical pain. This was an eye opening experience. Not long after, his wife became my client for bereavement counseling. I learned in more detail about the difficulty of her caring experience and her complex bereavement needs. At the time, I wondered why we couldn't have done more to help this couple. As a result of this experience, I realized the opportunity to perform dignity therapy with people who had MND would be challenging but it would also offer a substantial reward by providing an intervention that had the potential to alleviate distress in people with MND and their families.

### 1.5 Study background and funding

In 2009, Professor Samar Aoun of the Western Australian Centre for Cancer and Palliative Care (WACCPC) at Curtin University received a Linkage Grant (LP 0991305) from the Australian Research Council (ARC) to fund the current research. Professor Chochinov was a co-investigator on the grant. Linkage projects provide funding to support research projects that are collaborative, acquire new knowledge, and involve risk or innovation (Australian Research Council 2014). A partner organization must also make a financial contribution to the project, and in the case of this research, the Motor Neurone Disease Association of Western Australia (MNDAWA) was the partner. As part of the research costs, an Australian Postgraduate Award Industry (APAI) was awarded to provide a stipend to a PhD student to perform the research, which I received. The research commenced on 25 October 2010.

### 1.6 Aims of the study

Dignity therapy is an end of life psychotherapy and psychosocial intervention designed to enhance quality of life and moderate existential and psychosocial distress.

The objectives of this study were to assess the feasibility, acceptability, and potential effectiveness of dignity therapy for people with MND and their family carers.

The specific aims were to determine whether: a) dignity therapy increases hope, meaning, and dignity for people with MND; b) dignity therapy decreases perceived caregiver burden, anxiety and depression, and increases hope for MND family carers; c) dignity therapy is acceptable to people with MND and their family carers; and d) it is feasible to provide dignity therapy to people with MND.

## 1.7 Significance of the study

This study supports the growth and extension of palliative care research in relevant ways which directly address the three current challenges facing the provision of palliative care today. These challenges, as mentioned earlier, include providing psychosocial care; providing care and supports to family members; and providing palliative care to people with illnesses other than cancer. First, this study extends research and understanding in the area of psychosocial care at the end of life by advancing the research on dignity therapy. Next, this study is the first study to explore the impact of dignity therapy on family carers during their caring experience. Finally, this research examines a psychosocial intervention previously used with the traditional palliative care population – cancer patients – and extends it to a new palliative care group – people with MND, and it is the first study to examine the practical aspects of delivering psychosocial care to people with MND in light of the challenging physical symptoms often found in this population.

Moreover, this research responds to calls to develop and test psychosocial and spiritual interventions in the Report of the Quality Standards Subcommittee of the American Academy of Neurology 2009 ALS Practice Parameter update (Miller, Jackson et al. 2009) which has been endorsed by MND Australia (MND Care Net 2014). It also responds to the Palliative Care Australia's Standards on Providing Quality Palliative Care to All Australians by seeking to relieve suffering and improve the quality of living and dying through improved psychosocial care to people with life limiting conditions (Palliative Care Australia 2005).

## 1.8 Structure of the thesis

This thesis is presented in the form of a "hybrid" thesis by publication (Curtin University 2011 p. 5). In this format, the thesis is a typescript but some chapters are papers which have been published. Each chapter will begin with a brief introduction of what is contained within the chapter. A relevant quotation will also begin the chapter to provide a human voice and historical aspect to the thesis.

Chapter One has introduced and provided an overview of the research reported in the thesis. It has provided background information, including definitions for psychosocial and palliative care. This chapter has presented the three main challenges facing palliative care today, which include the provision of psychosocial care, the incorporation of care and supports for family carers of the terminally ill, and the inclusion of people facing death who have illnesses other than cancer. The researcher has been introduced and relevant experiences of the researcher were shared. The chapter concludes with the study background and funding, the aims and significance of the study, and the structure of the thesis.

Chapter Two describes the research setting and provides an overview of MND, including its diagnosis, symptoms and treatment. Best practice management is examined, including service provision in the research setting of WA. Key symptoms and management points along the MND trajectory related to psychosocial distress in people with MND and their family carers are explored.

Chapter Three is the literature review and is divided into three parts. The first section examines the psychosocial needs and stressors of people with MND. The second section examines the psychosocial needs of MND family carers. Because MND has no cure, maintaining quality of life for people with MND and their family carers is a primary treatment goal. Previous research has shown that quality of life for people with MND and their family carers is related to psychosocial and existential distress rather than physical impairment and decline. Despite this, psychosocial interventions have not been developed to treat distress. The third section provides a comprehensive review of the literature on dignity therapy, an end of life psychotherapeutic intervention designed

to alleviate existential distress, enhance quality of life, create spiritual peace, strengthen relationships, and increase meaning, purpose and dignity at the end of life.

Chapter Four is the research methodology. It is comprised of a paper entitled, "Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers?" This manuscript details the study protocol and was published in the peer-reviewed journal *BMC Palliative Care*. The design is a cross-sectional one group pre/post-test design. Outcomes for participants included hopefulness, spirituality/meaning and dignity. Outcomes for family carers included caregiver burden, hopefulness and anxiety/depression. Acceptability with both groups was to be determined through a feedback questionnaire, which was the same measure used in the original pilot study and the international randomized controlled trial (IRCT) of dignity therapy.

Chapters Five, Six, and Seven present the research findings. Chapter Five is a paper examining the impacts of dignity therapy on people with MND published in the interdisciplinary journal *PLoS One* entitled "Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease." The results of the study on 29 people with MND showed no significant changes on the outcome measures on the group level, but there were increases in hope on the individual level. Dignity therapy was acceptable and the results of the feedback questionnaire were similar to the results found in the IRCT. Dignity therapy is feasible if the therapist can overtime time and communication difficulties.

Chapter Six contains a paper examining the impacts of dignity therapy on MND family carers published in *BMC Palliative Care* entitled "Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neurone disease." The results showed no significant changes on the outcome measures on the group level, but there were some decreases in anxiety and depression in data analyzed on the individual level. These findings suggest dignity therapy may moderate or reduce anxiety and depression in distressed MND family carers. Family carers saw benefits from dignity therapy to people with MND and to themselves after bereavement, but acceptability was mixed at the time of the intervention and some family carers indicated

negative experiences from the therapy. Dignity therapy involving family carers is feasible and involvement of family carers had minimal impact on the therapy.

Findings on the feasibility of dignity therapy with people with MND and their family carers is included in Chapter Seven, specifically examining how common MND symptoms can affect the provision of dignity therapy and psychotherapeutic interventions more generally. The chapter also provides detail on the practical aspects of delivering dignity therapy, including the time and costs involved. Also contained in this chapter is a personal reflection related to feasibility issues and the length of time taken to perform dignity therapy which was published in the *Journal of Palliative Medicine* entitled "It takes the time that it takes." This chapter is presented in manuscript format with a brief introduction, details of the methodology, and a discussion to mirror the format used in Chapters Five and Six.

Chapter Eight is the discussion, which contains an overview of the key findings, a discussion of these findings in light of previous literature, implications for future research, strengths and limitations of the study, recommendations for clinical practice, and overall conclusion. The study's contribution to the three challenges in palliative care outlined earlier is discussed.

# CHAPTER TWO
# 2. Setting

Chapter Two provides an overview of MND, including diagnosis, physical symptoms and treatment. Best practice recommendations for the management of MND are examined, including service provision and challenges to service provision in the research setting of WA. Key symptoms and management points along the MND trajectory related to psychosocial distress in people with MND and their family carers are explored.

> I'm quite often asked, 'How do you feel about having ALS?' The answer is, 'Not a lot.' I try to lead as normal a life as possible, and not think about my condition, or regret the things it keeps me from doing, which are not that many. (Hawking 2008 at 0:22)

Stephen Hawking is a well-known British physicist who has written extensively on the origins of the Universe. Stephen Hawking has MND, is paralyzed and wheelchair-bound, has a tracheostomy and is dependent on a respirator to aid breathing, and communicates by using a computer with eye-gaze technology and a voice synthesizer.

## 2.1 Motor neurone disease

Motor neurone disease (MND), also called amyotrophic lateral sclerosis (ALS), is an incurable neurodegenerative disease. While MND and ALS are often used synonymously, use tends to vary by country with MND used in Australia and the UK, and ALS common in the US and Japan. MND is also sometimes called Lou Gehrig's disease in the US after a famous American baseball player who developed the disease. Motor neurones are specialized nerve cells that transmit information via electrical impulses from the brain and spinal cord to muscles resulting in movement (Kent 2012). When motor neurones are damaged, messages can no longer be carried by the nerve cells to the muscles. As a result, muscles become weak and atrophy. In MND, almost

all skeletal muscles are eventually affected (Kent 2012).

MND is always progressive with no periods of remission, although presenting symptoms and disease course vary between people (Mitsumoto and Rabkin 2007). A staging system for MND to help provide a universal and objective measure of disease progression has been developed in recent years, but research to validate this system is just beginning (Roche, Rosas-Garcia et al. 2012). As such, there is currently no staging system in MND with easily identifiable clinical milestones. On average, a person with MND will survive two to three years after being diagnosed (Mitchell and Borasio 2007, McDermott and Shaw 2008). Fifty percent of people with MND die within 14 months of diagnosis (Orrell 2010), and approximately 10% survive 10 years or more (Talbot, Turner et al. 2010). Noted theoretical physicist Stephen Hawking is an exceptional case, surviving over 50 years after being diagnosed at age 21 (Harmon 2012). Older age at symptom onset, early respiratory dysfunction, and bulbar-onset are associated with reduced survival, while limb-onset and younger age predict prolonged survival (Kiernan, Vucic et al. 2011). People with MND endure progressive paralysis and most die from respiratory failure, often precipitated by pneumonia and usually during sleep (McDermott and Shaw 2008, Kiernan, Vucic et al. 2011).

The cause of MND is unknown, although in 5-10% of cases there is a family history of the disease and a known genetic link (Mitchell and Borasio 2007). The mean age of onset is 59 years; however, the mean age of onset in people with familial MND is younger, from 47-52 (Andersen, Borasio et al. 2005). The age range of onset is from 18 to 90 (MND Australia 2010), but incidence decreases rapidly after age 80 (Kiernan, Vucic et al. 2011). Men are affected more often than women at a ratio of 1.6 to 1 (Borasio and Miller 2001, Mitchell and Borasio 2007), and whites are affected more than non-whites (Mitsumoto and Rabkin 2007). The annual incidence of the disease is 1.5 to 2 in 100,000, with an annual prevalence of about 7 per 100,000 population (Borasio and Miller 2001, McDermott and Shaw 2008) and a lifetime risk of 1 in 350-500 (Andersen, Abrahams et al. 2012). There is only one medication approved for treatment, Riluzole, which has numerous side effects and extends life by only a few months (Mitchell and Borasio 2007, Mitsumoto and Rabkin 2007). Recent MND research has focused on understanding the general pathophysiology, genetics, and

clinical phenotypes in MND in order to discover new treatment approaches (Kiernan, Vucic et al. 2011).

In Australia, it is estimated that 2 people per day are diagnosed with MND. On any one day, approximately 1,900 people have the disease. In 2011, 790 people in Australia died from MND (MND Australia 2014). The MND Association of Western Australia (MNDAWA) estimates that there are 100-120 people living with MND in Western Australia (WA), with 40 to 50 people dying from the disease each year. These numbers will increase as the population in WA increases and ages. At any one time, MNDAWA manages approximately 145 clients, including people with MND and families who are supported during bereavement (WA Department of Health 2008a).

### 2.1.1 Types of MND

MND is an overarching term for a group of disorders that affect motor neurones. There are four main groups which principally vary based on the location of the primary motor neurones affected.

- ALS – This form of MND has a spinal onset and involves both upper and lower motor neurones with weakness occurring in one or more limbs. People commonly present with weakness in the hands or feet, including tripping or grasping difficulties (Mitchell and Borasio 2007). ALS is the most common form of MND (Talbot, Turner et al. 2010).
- Progressive bulbar palsy – This form of MND has a bulbar onset and affects primarily the upper motor neurones. People commonly present with speech and swallowing issues, including slurred speech, choking, and difficulty chewing (Mitchell and Borasio 2007). A few will present with respiratory insufficiency, including breathlessness, poor sleep and morning headache (Kent 2012). Progressive bulbar palsy affects approximately 20-25% of those diagnosed with MND, and it has the shortest survival (McDermott and Shaw 2008, Kent 2012).
- Progressive muscular atrophy – This form of MND primarily affects the lower motor neurones. People commonly present with weakness in the

legs. People with this presentation typically survive longer, from five to ten years from symptom onset (Kent 2012).

- Primary lateral sclerosis – This form of MND only affects the upper motor neurones. It is the least common form, affecting 3-5% of people with MND, and typically progresses more slowly than ALS (Kiernan, Vucic et al. 2011).

Though each group may present and progress differently, there is significant overlap of symptoms among the different types. Eventually, these different groups may develop symptoms in all areas of the body (Oliver and Aoun 2013).

### 2.1.2 Physical Symptoms

Direct symptoms resulting from the degeneration of motor neurones include muscle weakness and atrophy, twitching, muscle cramps, speech difficulty (dysarthria), shortness of breath (dyspnea), difficulty swallowing (dysphagia), and uncontrolled crying or laughter (emotional lability/pseudobulbar affect) (Oliver and Borasio 2004, Mitchell and Borasio 2007). Indirect symptoms include pain, drooling (sialorrhea), thick mucus secretions, constipation, sleep problems, and psychological disturbances (Mitchell and Borasio 2007). There is also an association between frontotemporal dementia (FTD) and MND (Lomen-Hoerth, Murphy et al. 2003, Kiernan, Vucic et al. 2011), and some level of cognitive decline is encountered in approximately 50% of people with MND (Merrilees, Klapper et al. 2010).

### 2.1.3 Management

Intervention for MND centers on managing symptoms and the provision of palliative care as there are limited options for treatment and no cure (Miller, Jackson et al. 2009). Care goals include maintaining independence and physical function for as long as possible, managing physical symptoms, facilitating communication about end of life issues, and providing psychosocial support for people with MND and their family (Kristjanson, Toye et al. 2003, Oliver and Borasio 2004, Mitsumoto, Bromberg et al. 2005, Kent 2012). It is recommended that people with MND receive coordinated and comprehensive care at a multidisciplinary clinic from a team which may include a

neurologist, gastroenterologist, rehabilitation specialist, physiotherapist, occupational therapist, respiratory therapist, speech therapist, dietician, social worker, research coordinator, mental health professional, neuropsychologist, nurse case manager, and palliative care physician (Miller, Rosenberg et al. 1999, Mitsumoto and Rabkin 2007, Andersen, Abrahams et al. 2012). Multidisciplinary care received through specialized MND clinics improves quality of life and lengthens survival (Van den Berg, Kalmijn et al. 2005, Miller, Jackson et al. 2009). Attendance at clinics also reduces hospital admissions (Andersen, Borasio et al. 2005). Good communication between health care providers is essential, and the care received at multidisciplinary clinics is typically coordinated with care received from general practitioners and community-based services (Andersen, Borasio et al. 2005, Kent 2012).

Depending on disease progression, most people with MND will confront numerous decisions regarding medical interventions to manage their increasing impairment and decline in function. Such interventions include: the use of mobility aids such as walkers, wheelchairs, and hoists; the insertion of a gastronomy tube or percutaneous endoscopic gastronomy (PEG) to aid nutrition; the use of assisted ventilation to aid breathing when respiratory muscles weaken; and the use of augmentative and alternative communication (AAC) devices to supplement or replace loss of speech. Because muscle weakness in the lower limbs can result in falls and impaired walking, people with MND may require a wheelchair for safe mobility (Kent 2012), modifications to their homes, and/or other mobility aids. People with MND who have impaired swallowing may desire a PEG tube to aid eating and drinking, which is the standard procedure (Leigh, S Abrahams et al. 2003). Insertion of a PEG can prolong life, as malnutrition and weight loss are associated with shortened survival (McDermott and Shaw 2008). Weakness of respiratory muscles can cause numerous problems, including dyspnoea, anxiety, decreased clearance of saliva and respiratory infections (Kent 2012). Non-invasive ventilation (NIV), which is ventilation that supports breathing through a mask rather than through a breathing tube, improves symptoms caused by respiratory insufficiency, improves quality of life, and increases survival (Miller, Jackson et al. 2009). In addition, people with MND may encounter speech impairment or lose the ability to speak altogether. In bulbar presentation, this can occur

quite early in the disease trajectory. Speech therapy and the transition to AAC can help maintain the ability to communicate (Brownlee and Bruening 2012).

#### 2.1.3.1 Local context

In Australia, a General Practitioner (GP) is usually the primary health professional and the first point of contact for a person with MND. Most states and regions, with the exception of Tasmania, the Australian Capital Territory (ACT), and the Northern Territory, have multidisciplinary clinics located in major metropolitan hospitals. In Tasmania, a regional advisor performs assessments and makes appropriate referrals for the care of people with MND. The ACT is served by the MND Association and three multidisciplinary clinics in adjacent New South Wales. The Northern Territory does not have a multidisciplinary clinic nor a motor neurone disease association, and people with MND in that state are referred to the MND Association in New South Wales for advice and assistance (MND Care Net 2014).

In WA, the geographical area focused on in this study, there are two specialised hospital-based multidisciplinary MND clinics in the Perth metropolitan region that provide care to people with MND. There is also a satellite clinic in Bunbury, two hours south of Perth. People with MND are typically scheduled to attend the clinic every three months. In WA, about 20% will choose a form of feeding support like a PEG and about 20% of people with MND receive NIV and (WA Department of Health 2008a). People in WA use assisted ventilation more frequently than those in Europe (10%) but less frequently than those in Japan (46%) (Borasio, Gelinas et al. 1998, Leigh, S Abrahams et al. 2003).

People with MND also typically visit their GP and receive social support, care advice and advocacy from the MNDAWA. For people under the age of 65, the Multiple Sclerosis Society of WA provides in-home care and respite for people diagnosed with rapidly degenerative neurological conditions, including MND, through the Neurological Conditions Coordinated Care Program (NCCCP) (Bahn and Giles 2012). People over the age of 65 can access in-home or residential care through government aged-care packages. To access these services, a member of the aged care assessment team (ACAT) within the Department of Health performs an assessment and makes a referral

for the appropriate level of service. Services range from supportive services, such as personal care, meals and nursing visits to help a person stay at home, to 24-hour high-level care in a residential facility. Additionally, community palliative care is available at the end-stage by referral from a physician. Despite these services, there is a lack of coordination between services for people with MND, as well as gaps in respiratory specialist support and allied health support, including social work, counseling, and psychological care (WA Department of Health 2008a).

Formal psychosocial care can be accessed through a GP, who can provide a referral for mental health services provided by GPs, psychologists, psychiatrists, and eligible social workers. The Better Access to Mental Health program provides Medicare rebates for a maximum of 10 individual and 10 group sessions per year (Department of Health 2012). NCCCP and community palliative care both employ counselors, who can provide services on request. MNDAWA Care Advisors also provide informal psychosocial support (WA Department of Health 2008a).

Service delivery in WA is complicated by its large geographical size and population dispersion. WA is Australia's biggest state, accounting for 33% of the Australian continent and covering an area of 2,525,500 $km^2$. WA is bigger in size than Western Europe and four times the size of Texas. Meanwhile, WA's population of 2.5 million represents a population density of only .8 people per square kilometre and accounts for only 11% of the national total. In comparison, the population density of the US is 32 people per square kilometre, whereas the whole of Australia is 8 people per square kilometre. Approximately 80% of WA's population lives in the Perth metropolitan area; however, 500,000 people reside in rural and remote areas where accessing services can be difficult (Australian Bureau of Statistics 2013). Figures from WA by region from 2001 through 2006 reveal that 19% of people with MND were treated by WA County Health Service in rural areas, with 81% treated in the Perth metropolitan area (WA Department of Health 2008a). People in rural areas are served by their GPs and are unlikely to receive MND specialist input into their disease management. The lack of neurologist specialist care and allied supports in rural and regional areas presents a dilemma in the equity of care for people with MND and their families for which there is no easy answer (Smith 2007, WA Department of Health

2008a).

### 2.1.4 Key symptoms and management points along the MND care trajectory related to psychosocial distress in people with MND

The following symptoms and management points in MND care during the disease course are related to psychosocial distress and should be considered in the provision of psychosocial care for people with MND and their families. (The psychosocial needs of people with MND will be discussed in more detail in Chapter 3).

#### 2.1.4.1 Diagnosis

Diagnosis of MND in the early stages of the disease is difficult. The average delay from onset of symptoms to diagnosis is 14 months (Leigh, S Abrahams et al. 2003), this is in part due to their being no definitive diagnostic test. Diagnostic criteria (called the El Escorial Diagnostic Criteria) require the following to be present: signs in more than 1 of 4 body regions; evidence of both upper and lower motor neurone degeneration; and progressive spread (Mitchell and Borasio 2007, Mitsumoto and Rabkin 2007). In addition, because MND is a rare disease, most GPs will see only one or two cases in their careers which contributes to the difficulty of formulating the diagnosis (McDermott and Shaw 2008). An accurate and early diagnosis is important because delay may cause anxiety and impair a person's social and professional life (Andersen, Borasio et al. 2005). Uncertainty about the diagnosis can cause fear, confusion and worry, and these feelings are amplified when experienced over a long period of time, resulting in psychological distress (O'Brien, Whitehead et al. 2011a, Mistry and Simpson 2013).

When a diagnosis is made, it is experienced as a shocking and devastating experience for people with MND and their family members, and some will need psychological support to help them cope (Borasio, Sloan et al. 1998, WA Department of Health 2008a, Andersen, Abrahams et al. 2012, Mistry and Simpson 2013). Many will undergo a period of reactive depression and may benefit from psychological intervention (Mitchell and Borasio 2007). The way the diagnosis is communicated can have a profound effect on people with MND and their family members (O'Brien, Whitehead et al. 2011a, Andersen, Abrahams et al. 2012), yet surveys have shown that people with

MND and their family members perceive that the diagnosis is communicated in a less than satisfactory manner 50% of the time (Borasio, Sloan et al. 1998, McCluskey, Casarett et al. 2004). When the diagnosis is communicated poorly, it can leave people with MND feeling hopeless and abandoned, and negatively impact their relationship with their physician (Andersen, Borasio et al. 2005, O'Brien, Whitehead et al. 2011a). Insensitive delivery of the diagnosis can also affect psychological adjustment to loss and grief (Ackerman and Oliver 1997, Borasio, Sloan et al. 1998). At the time of diagnosis, clinical management guidelines highlight the need to provide assurance of ongoing support by a multidisciplinary team (Andersen, Borasio et al. 2005, Andersen, Abrahams et al. 2012), including psychological support and counseling (Leigh, S Abrahams et al. 2003). Nonetheless, research findings suggest people with MND do not receive sufficient emotional support at the time of diagnosis (O'Brien, Whitehead et al. 2011a).

#### 2.1.4.2 Loss of speech

Verbal communication is impaired in 70-80% of people with MND during the course of the disease (Leigh, S Abrahams et al. 2003, Oliver and Borasio 2004). In people with bulbar onset, problems with speech are an early symptom which usually progresses to speech loss within months (Leigh, S Abrahams et al. 2003). Speech loss and impairment can adversely affect the quality of life of people with MND and their family members (Murphy 2004) because communication is fundamental to social closeness and participation in life (Miller, Rosenberg et al. 1999). To limit impairment, early involvement of a speech therapist is recommended to allow time to plan and adapt to AAC strategies and aids (Oliver and Borasio 2004). AAC aids and devices include unaided (gestures, signs, facial expressions) low-tech (white board, or letter or picture boards), and high tech aids (computer-based systems) (Brownlee and Bruening 2012). AAC can improve quality of life for people with MND by maintaining function, offering a greater sense of independence, providing the means to assist with medical decision making, improving relationships, and providing opportunities for social support and personal growth (Brownlee and Palovcak 2007, Joubert 2013). The overall goal in this area is for health care providers is to provide supports to optimise meaningful interpersonal communication (Andersen, Abrahams et al. 2012).

In WA, there are speech therapists associated with the MND multidisciplinary

clinics, and referral can be made to AAC specialists at the Communications and Assistive Technology Service, Department of Medical Engineering and Physics, Royal Perth Hospital, who have knowledge of the full range of AAC aids. With these resources, people with MND are able to select technologies depending on their needs, abilities and financial resources, ranging from low tech white boards to various methods of computerized type-to-speech technology, including tablet computers and Lightwriters, which are small and portable type-input voice-output devices.

#### 2.1.4.3 Emotional lability

Emotional lability, also called pseudobulbar affect, is experienced by up to 50% of people with MND (Mitchell and Borasio 2007). A person experiencing emotional lability will display an affect triggered by an emotion that is discordant to the situation, such as mood-incongruent, inappropriate, or uncontrolled crying or laughing (Elman, Houghton et al. 2007). For example, a nostalgic memory which may evoke a sigh in most people can evoke a prolonged crying episode in someone experiencing emotional lability. It is not a mood disorder or mental illness (Elman, Houghton et al. 2007, Mitchell and Borasio 2007), rather it is caused by the neurodegenerative disease affecting the brain, and a person experiencing this symptom has little control (Andersen, Abrahams et al. 2012). It can be very disturbing to the person with MND and others in social situations (Mitchell and Borasio 2007), and in severe cases may be disabling, primarily due to the stigma attached to losing emotional control (Moore, Gresham et al. 1997). There are drug treatments (Miller, Jackson et al. 2009, Kent 2012), but only 15% of people with MND request these drugs (Meininger 2005) despite recommendations that emotional lability receive as much attention as other physical symptoms in MND (Goldstein, Atkins et al. 2006). People with MND and their families can find this symptom distressing but are often unaware it is a symptom of MND (Elman, Houghton et al. 2007). Emotional lability in the person with MND is a predictor of psychological distress in MND family carers (Goldstein, Atkins et al. 2006), thus treatment may benefit both the person with MND and their family carer. Emotional lability has the potential to cause distress and interfere with psychotherapy where triggering emotions may be evoked, especially in people with MND who are unaware of this symptom.

#### 2.1.4.4 Cognitive and neurobehavioral decline

For many years, people diagnosed with MND were told their brain remained untouched while their body deteriorated (Kent 2012). Today, it is understood that there is an overlap between MND and frontotemporal lobe degeneration which may lead to frontotemporal dementia (FTD) (Phukan, Pender et al. 2007). Impaired frontal lobe function has been reported in up to 50% of people with MND (Merrilees, Klapper et al. 2010, Raaphorst, De Visser et al. 2010). Approximately 15% of people with MND will meet the criteria for FTD (Ringholz, Appel et al. 2005, Miller, Jackson et al. 2009, Merrilees, Klapper et al. 2010) with 11% of that total meeting the criteria for the behavioural variant of FTD, characterised by apathy, disinhibition, irritability and lack of empathy (Lillo, Mioshi et al. 2011). FTD and cognitive-behavioral decline are more common in people with bulbar presentation (Achi and Rudnicki 2012), and in some, FTD is the presenting symptom of MND (Elamin, Phukan et al. 2011). To date, there are no empirically tested treatments available for cognitive or behavioral impairment in MND (Miller, Jackson et al. 2009).

FTD manifests symptoms in three areas: language, behavior and/or personality, which may overlap and exist on a continuum (Achi and Rudnicki 2012). In some people with MND, cognitive impairment results in speech and language dysfunction (Leigh, S Abrahams et al. 2003, Giordana, Ferrero et al. 2011). Other people will experience a change in behaviour: apathy, executive dysfunction and disinhibition are the most commonly cited neurobehavioral symptoms of people with MND (Chio, Vignola et al. 2010, Merrilees, Klapper et al. 2010, Lillo, Mioshi et al. 2011). People with MND who develop FTD may exhibit severe symptoms including aggression, irritability, impulsivity, loss of insight, self-centeredness, lack of empathy, food cravings, and obsessive-compulsive behaviors (Merrilees, Klapper et al. 2010).

A comprehensive neuropsychological assessment is recommended according to MND management guidelines (Phukan, Pender et al. 2007, Miller, Jackson et al. 2009) but is not feasible for most clinics who have inadequate resources (Achi and Rudnicki 2012). Common dementia screenings like the Mini-Mental State Examination are insensitive to cognitive and behavioral impairment in people with MND (Andersen,

Abrahams et al. 2012). Short screening tests specifically for people with MND have been developed, such as the Addenbrooke's Cognitive Examination Revised (ACE-R) (Mioshi, Dawson et al. 2006) and the ALS Cognitive Behavioral Screen (Woolley, York et al. 2010) but are not yet in widespread use (Bede, Oliver et al. 2011). In WA, neuropsychological assessments are very rarely completed and cognitive screening measures are not used (A. Kermode, personal communication, 1 Oct. 2011).

Cognitive-behavioral symptoms can have a significant impact on people with MND and their families, and negatively affect quality of life, including relationships, social supports, and adherence with treatment (Bede, Oliver et al. 2011, Lillo, Mioshi et al. 2011). Practice guidelines suggest that healthcare professionals become knowledgeable about cognitive behavioral impairment in MND and pursue training in the management of this symptom (Andersen, Abrahams et al. 2012). Potentially, concepts learned from the management of dementia can be applied to people with MND who have impaired frontal lobe function.

#### 2.1.4.5 End of life care

The current suggested clinical approach for MND advocates that palliative care should begin at the time of diagnosis rather than in the final weeks of life (Leigh, S Abrahams et al. 2003, Mitsumoto, Bromberg et al. 2005, Andersen, Borasio et al. 2007, Andersen, Abrahams et al. 2012), but there are no accepted international standards for when palliative care should be initiated (Bede, Oliver et al. 2011). Late referral of people with MND to palliative care services is common (Bede, Oliver et al. 2011). In the US, for example, strict criteria by Medicare for access to palliative care services results in many people with MND dying without adequate access to end of life care (Elman, Houghton et al. 2007). In a European survey of specialist MND care providers it was reported that the available medical care for people with MND was insufficient in the terminal phase in 33% of cases from Northern Europe and 61% of cases from Southern Europe (Borasio, Shaw et al. 2001). In separate research with service users in the UK and Australia, people with MND and their families reported they found it difficult to access palliative care services (Ray and Street 2011, O'Brien, Whitehead et al. 2011b).

In the Australian state of Victoria, the Department of Health commissioned a study called the MND Pathway Project citing a need to assist people with MND access palliative care services as well as a need to assist palliative care services manage and support people with MND (Victorian Government Department of Human Services 2008). An interim report determined that palliative care of people with MND could not be integrated into existing frameworks, that palliative care workers were not confident in their knowledge about MND, and people with MND lacked understanding about palliative care services (Victorian Government Department of Human Services 2008). Between 2000 and 2002, less than 10% of people in WA who died of non-malignant diseases accessed palliative care services, compared with 66% of people who died of cancer (McNamara, Rosenwax et al. 2006). This research demonstrates a clear need for increased provision of palliative care services for people with MND.

Palliative care offers important services at the end of life that are not a primary focus of MND multidisciplinary care. These services include terminal phase pain and symptom management, the provision of practical and emotional supports for MND caregivers such as respite care and counseling, preparation of advance directives, assistance with end of life decision making, and the provision of information about death (Bede, Oliver et al. 2011). It is argued that without these services, many of the needs of people with MND and their family carers will not be adequately met. Palliative care, which provides support for psychosocial and existential distress, is especially important for people with MND as research indicates feelings of hopelessness and being a burden on family can lead to requests for a hastened death or suicide (Ganzini, Johnston et al. 1998, Ganzini, Silveira et al. 2002).

## 2.2 Family caregiving in MND

Generally, end of life caregiving by families is demanding and associated with psychological distress (Leblanc, London et al. 1997, Pinquart and Sorenson 2003, Aoun, Kristjanson et al. 2005). In MND, the demands are greater than most other diseases, and the burden of the disease falls heavily on family carers (Hecht, Graesel et al. 2003). MND family carers experience exceptional strain due to the rapid and progressive nature of the disease, coupled with the incapacitating effects of MND (Chio, Gauthier et al.

2005). In many ways, care for a person with MND is similar to family caregiving in other terminal disease populations. However, there are also unique features in MND family caregiving due to the disease trajectory of MND (Oliver and Turner 2010), the particular challenges faced by MND family carers, and the lack of available supports to MND family carers (Grande, Stajduhar et al. 2009). Family carers of terminally ill people in general have numerous unmet psychosocial needs (Hudson and Payne 2011, Harding, List et al. 2012), and this is mirrored in MND. MND practice guidelines state that the provision of psychosocial care for MND family carers is a vital area for research (Mitsumoto, Bromberg et al. 2005), and there are calls to develop psychosocial interventions for MND family carers (Aoun, Bentley et al. 2013).

Australian statistics indicate that most of the daily care for people with MND is provided by family carers throughout the course of illness (Love, Street et al. 2005). MND family carers encounter significant life changes that include both daily changes and long term adjustments (Hughes, Sinha et al. 2005). They provide an enormous amount of care which grows over time as people with MND require increasing assistance with nearly all aspects of daily living (Chio, Gauthier et al. 2005). Time spent caring for people with MND is related to overall disability and ranges from 5 hours per day for those with mild disability to 15 hours per day for those with severe impairment (Chio, Gauthier et al. 2006). A progressive increase occurs in time spent on bathing, dressing, toileting, administering medications, and feeding, corresponding with level of disability (Chio, Gauthier et al. 2006). MND family carers also face anxieties about illness progression, the well-being of family (Bromberg and Forshew 2002, Trail, Nelson et al. 2004); changes in family roles (Lackey and Gates 2001, Hughes, Sinha et al. 2005); loss of a reciprocal relationship (Ray and Street 2006b); and being judged and blamed by other family members for care decisions (Martin and Turnbull 2001).

People with MND in WA will meet an average of 82 different health care providers during the course of the disease (WA Department of Health 2008a), further increasing the burden on carers who manage visits and coordinate appointments with other family commitments. The time demands on Australian MND family carers and the lack of respite care results in feelings that the carer's life is on hold and completely occupied with their caring role (Kristjanson, Toye et al. 2003). Family carers in rural

areas are further disadvantaged due to the lack of supports such as in-home care and respite care (Bahn and Giles 2012).

MNDAWA provides psychosocial support to carers through carer training sessions and a Carer Support Program, which includes monthly educational meetings and peer support. In addition to their other roles, MND care advisors offer informal counseling and psychosocial care to MND family carers, and they also provide bereavement support for up to six months after death; however, the small number of care advisors employed by MNDAWA limits their availability (WA Department of Health 2008a). There is a lack of equitable access for people with MND to palliative care services in WA (WA Department of Health 2008a, WA Department of Health 2008b), and as a result many family carers miss out on the psychosocial and supportive care provided by palliative care at the end of life.

### 2.2.1 Key symptoms and management points along the MND care trajectory related to psychosocial distress in MND family carers

The following symptoms and management points in MND care are related to psychosocial distress for MND family carers and should be considered in the provision of psychosocial care for people with MND and their families. (The psychosocial needs of MND family carers will be discussed in more detail in Chapter 3).

#### 2.2.1.1 Cognitive and neurobehavioral decline

Cognitive and behavioral changes associated with MND can have a profound impact on family carers, sharply increasing depression and burden and negatively affecting their quality of life (Chio, Vignola et al. 2010, Merrilees, Klapper et al. 2010). Some family carers observe these changes, while the person with MND may lack insight and be unaware (Woolley, Moore et al. 2010). Other family carers fail to recognize symptoms that develop slowly, and some use denial, where they negate and dismiss observable symptoms, in order to cope (Woolley, Moore et al. 2010). Determining the full impact of FTD in people with MND on their family carers is not yet known and requires further investigation (Lillo, Mioshi et al. 2011). Recommended screening methods and procedures are in development (Mioshi, Dawson et al. 2006, Woolley, York et al. 2010), but robust and validated interventions to support carers are lacking.

#### 2.2.1.2 Assisted ventilation

Respiratory muscle weakness increases over time, and people with MND begin to experience breathlessness (Mustfa, Walsh et al. 2006). Breathlessness and subsequent assisted ventilation can cause family carers to feel helpless and ill-prepared to cope, causing anxiety, depression, and distress (Gysels and Higginson 2009). Assisted ventilation increases survival time for people with MND, which leads to an extended period of caregiving (Mustfa, Walsh et al. 2006, Rabkin, Albert et al. 2006). When a person with MND is ventilator-dependent, family carers often feel excessively burdened and report feelings of resentment, frustration and unhappiness (Gelinas, O'Connor et al. 1998). Continuous ventilator use can also result in a person with MND losing the ability to communicate (Kent 2012), and sexuality is negatively affected (Kaub-Wittemer, Steinbuchel et al. 2003), all of which can add to the stress and burden on MND carers.

NIV, which involves positive-pressure ventilation through a face mask which is often used only at night (McDermott and Shaw 2008), is the preferred therapy for respiratory insufficiency (Andersen, Abrahams et al. 2012). The impact of NIV itself on caregiver burden is minimal (Baxter, Baird et al. 2013); however, it can create stressful and complex issues at the end of life with decisions around whether and when it should be withdrawn (Oliver and Turner 2010). Long term mechanical ventilation (LTMV), where breathing is performed continuously through an intubation tube, is used less frequently and is not recommended due to its high cost and the substantial emotional and social impacts on both people with MND and their caregivers (Andersen, Abrahams et al. 2012). LTMV often results in 'locked-in syndrome' where the person with MND becomes completely paralyzed and unable to communicate (Leigh, S Abrahams et al. 2003). LTMV can inadvertently result when people with MND present to an emergency room in respiratory failure (Eng 2006). These patients are rarely able to wean off the ventilator without causing death, creating a situation where both options, including continuing to live dependent on a ventilator or withdrawal of the ventilator and subsequent respiratory failure and death, are highly distressful (Leigh, S Abrahams et al.

2003, Eng 2006). Several studies have shown that caregiving for people with MND using LTMV increases burden and decreases quality of life for the family carer (Kaub-Wittemer, Steinbuchel et al. 2003, Akiyama, Kayama et al. 2006, Rabkin, Albert et al. 2006). The use of LTMV varies between countries (Andersen, Abrahams et al. 2012). It is often used in Japan, sometimes used in the US, and rarely used in the UK and Australia (Borasio, Gelinas et al. 1998).

#### 2.2.1.3 End of life caregiving

MND family carers gradually lose the ability to interact with their loved ones at the end of life, causing them to feel powerless and struggle with meaning (Akiyama, Kayama et al. 2006). Important components of the relationship deteriorate, such as sex life and marital communication, which present additional psychosocial and emotional challenges for spouse carers (Wasner, Bold et al. 2004, O'Connor, McCabe et al. 2008, Atkins, Brown et al. 2010). As people with MND lose function, family carers experience increased symptoms of somatic depression, including reduced quality of sleep and appetite (Pagnini, Rossi et al. 2010). Conflict can be created when people with MND and their family have different beliefs or attitudes about care (Bolmsjo and Hermeren 2003). People with MND may refuse life-sustaining treatments, such as insertion of a PEG tube or use of assisted ventilation (Kaub-Wittemer, Steinbuchel et al. 2003), or they may have an interest in hastening death, which can be upsetting for family carers (Rabkin, Wagner et al. 2000).

#### 2.2.1.4 Bereavement

Caregiving ends at death but MND continues to affect families. During bereavement, MND family carers experience lasting emotional effects, including sadness, anger, fear, frustration, depression, and, for some, hopelessness (Hebert, Lacomis et al. 2005). MND practice guidelines suggest spiritual and bereavement care be provided to family members as a way of preventing complicated bereavement (Mitchell and Borasio 2007). However, in a Canadian study where 27 bereaved MND family carers were surveyed, 37% of caregivers reported that they were coping poorly or very poorly sometimes years after death, and 68% reported a need for ongoing support (Martin and Turnbull 2001). A Western Australian study revealed that MND family

carers may be more vulnerable to prolonged grief and suggests that those who had delayed access to palliative care services have the at highest risk (Aoun, Connors et al. 2012). While there is recognition of the value of grief and bereavement support for MND family carers in the literature, this support is often reported to be inadequate by both MND family carers and MND specialist health care providers (Hebert, Lacomis et al. 2005, Aoun, Connors et al. 2012).

## 2.3 Summary

MND is a relatively rare neurodegenerative disease with no cure. People with MND gradually lose function of all voluntary muscles and become paralyzed. Most people with MND die from respiratory failure. Treatment centers on managing symptoms and providing palliative care, with goals of maintaining function and quality of life for as long as possible, and providing psychosocial support to the person with MND and his/her family. During the course of the disease, several symptoms and management points are related to psychosocial distress and the provision of psychosocial care. These include diagnosis, speech loss, emotional lability, cognitive and neurobehavioral decline, and end of life care. Optimal care is provided by MND multidisciplinary clinics working together with GPs, palliative care, and community-based services.

MND family carers experience significant strain during the caring experience. They provide long hours of care and face numerous anxieties. Management points in the care trajectory of a person with MND that relate to psychosocial distress in MND caregivers are cognitive and neurobehavioral decline, the use of assisted ventilation, end of life caregiving, and bereavement.

There are 100-120 people living with MND in WA, and 40-50 die each year. Multidisciplinary care is provided at two hospital-based MND clinics in the Perth metropolitan area and a satellite clinic in Bunbury. People with MND are typically scheduled to attend the clinic every three months. People with MND also typically visit their GP and receive social support, care and advocacy from MNDAWA. For people under the age of 65, the Multiple Sclerosis Society manages a program called NCCCP to provide in-home and respite care for people with MND, including home visits from a

neurological nurse. People over the age of 65 can apply for in-home care available to older people, although this does not include the specialist services of a neurological nurse. Community palliative care may be available upon referral from a physician, and research has shown people with MND have difficulty accessing these services in WA.

WA covers a vast area of 2,525,000 $km^2$ and its size is a challenge for service provision. About 19% of people with MND in WA live in rural areas, where a lack of neurologist specialist care and allied supports is common. The current situation presents an inequity in care available to people with MND and their families who live in rural and remote areas.

Neuropsychological assessments are extremely rare in MND care in WA. Because FTD may not be adequately screened for and treated in WA, MND family carers and health care providers may not be aware of cognitive and neurobehavioral decline, and there are missed opportunities to provide support in this important area. Psychosocial care can be accessed by referral from a GP. NCCCP and community palliative care employ counselors and social workers who can provide psychosocial support to people with MND and their families. MNDAWA Care Advisors also provide informal psychosocial support, which extends into bereavement for family carers. Overall, people with MND and their family carers are presented with numerous stresses and challenges over the course of the disease and their psychosocial needs are largely unmet.

# CHAPTER THREE
## 3. Literature Review

Chapter Three presents a review of the literature and it is divided into three sections. The first section examines the psychosocial needs and stressors of people with MND. The second section examines the psychosocial needs and stressors of MND family carers. Together, these two sections present evidence that maintaining quality of life for people with MND and their family carers is a primary treatment goal. Previous research has shown that quality of life for people with MND and their family carers is dependent on psychosocial and existential issues, rather than physical decline. Despite this, no psychosocial interventions have been developed to treat distress and improve quality of life. The third section provides a comprehensive review of the literature on dignity therapy, a palliative care psychosocial intervention designed to alleviate existential distress, enhance quality of life, and increase meaning, purpose and dignity at the end of life.

Fans, for the past two weeks you have been reading about a bad break I got. Yet today I consider myself the luckiest man on the face of the earth. I have been in ballparks for seventeen years and have never received anything but kindness and encouragement from you fans.

Look at these grand men. Which of you wouldn't consider it the highlight of his career to associate with them for even one day?

Sure, I'm lucky. Who wouldn't consider it an honor to have known Jacob Ruppert? Also the builder of baseball's greatest empire, Ed Barrow? To have spent the next nine years with that wonderful little fellow Miller Huggins? Then to have spent the next nine years with that outstanding leader, that smart student of psychology, the best manager in baseball today, Joe McCarthy!

Sure, I'm lucky. When the New York Giants, a team you would give your right arm to beat, and vice versa, sends you a gift -- that's something!

> When everybody down to the groundskeepers and those boys in white coats remember you with trophies -- that's something.
>
> When you have a wonderful mother-in-law who takes sides with you in squabbles against her own daughter -- that's something. When you have a father and mother who work all their lives so that you can have an education and build your body -- it's a blessing! When you have a wife who has been a tower of strength and shown more courage than you dreamed existed-- that's the finest I know.
>
> So I close in saying that I might have had a tough break, but I have an awful lot to live for! (Gehrig 1939 para. 3).

Lou Gehrig was a celebrated first baseman for the New York Yankees baseball team. He gave the above farewell speech to 60,000 fans at Yankee Stadium on 4 July 1939, confirming his diagnosis of MND/ALS, which came to be known thereafter as Lou Gehrig's disease in the US. He died less than two years later at age 37. This well-known speech highlights the importance of meaning, hope, and relationships in coping with MND.

## 3.1 Introduction

MND presents numerous psychosocial stressors and challenges for a person diagnosed with the disease and their family. The literature presented in this chapter articulates these challenges and includes findings from empirical research on issues relating to quality of life. Understanding quality of life is necessary in order to address the total pain (Saunders 1964) of people with MND and their families and carry out Dame Cicely Saunders' grounding philosophy that says palliative care should help people with life limiting illness live fully until death (Saunders 1976). Through the literature review, characteristics of interventions that may benefit people with MND and their family carers are revealed, which lays the groundwork for understanding why dignity therapy may prove beneficial.

This section also reviews the literature on dignity therapy. Dignity therapy is based on research that produced a theoretical model of dignity in the terminally ill. Using this model, dignity therapy was created, trialed and tested in a palliative care

population where almost all participants had cancer. After the pilot testing showed promising benefits for both people with terminal disease and their families, an IRCT with people with end-stage cancer was performed demonstrating similar benefits. Dozens of studies have followed exploring dignity therapy in more detail, including effectiveness studies, feasibility studies with different groups, cultural studies, case studies, studies exploring the themes found in dignity therapy documents, clinical perspectives, and implementation studies. A review of the resulting published literature will aid in understanding the possible benefits and limitations of dignity therapy for people who have MND.

## 3.2 Psychosocial Needs of People with MND

### 3.2.1 Quality of life

The WHO defines quality of life (QOL) as "individuals' perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (World Health Organization 1997). More simply, QOL can be described as the degree of similarity between a person's hopes and expectations and their current reality (Calman 1984). It encompasses bio-psycho-social-spiritual issues encountered during illness, including biological aspects (symptoms), psychological aspects (thoughts and emotions), social aspects (relationships, resources and supports), and spiritual aspects (religious and existential concerns) (Pagnini, Rossi et al. 2010). In MND, because there is no curative treatment, care is focused on maintaining QOL. While there appears to be a determined community of researchers examining QOL and the psychosocial aspects of MND, the research published in this area represents only about 8% of the total articles published relating to the illness (Pagnini, Simmons et al. 2012). The majority of published research continues to be conducted in the biological realm, typically on identifying causes and ameliorating physical symptoms.

Despite this focus, research has shown that QOL in people with MND is more likely to be related to psychosocial and spiritual conditions of life such as social support, hopelessness, meaning and purpose, and sense of burden, rather than biological aspects and physical condition (Ganzini, Johnston et al. 1999). In a prospective study with 96

participants, Simmons et al. (2000) found psychosocial and existential concerns were significantly correlated to QOL in people with MND, whereas physical impairment was not. Clarke (2001) confirmed these results and acknowledged the significant relationship between QOL and psychosocial wellbeing, particularly in people with MND with the greatest physical disability. A recent retrospective study of 72 people with MND further validated that QOL in people with MND is independent of physical function and that QOL does not decline as physical function declines (Cupp, Simmons et al. 2011).

Research has further determined that QOL in people with MND is related to several psychosocial, existential and psychological stressors. For example, Gibbons et al. (2013) conducted a quantitative study with 147 participants in the UK and found the psychosocial variables related to QOL in people with MND are depression and social withdrawal. Several QOL studies have shown that people with MND view social and spiritual areas as the most important components of QOL (Chio, Gauthier et al. 2004, Neudert, Wasner et al. 2004, Foley, O'Mahony et al. 2007, Roach and Averill 2009). In a quantitative study with 80 participants in Italy, Chio and colleagues (2004) found QOL was related to supportive and spiritual factors, while a German study with 51 participants found family and friends have the strongest effect on QOL (Neudert, Wasner et al. 2004). In an Irish qualitative study with five purposively selected participants, all participants placed importance on their faith in helping to maintain QOL (Foley, O'Mahony et al. 2007). Thus, psychosocial and spiritual health emerges as the most important factor affecting QOL. In addition, emotional wellbeing is associated with longer survival in people with MND based on a longitudinal study with 144 participants in the US (McDonald, Wiedenfeld et al. 1994). Nonetheless, the physical aspects of MND continue to receive the most attention in MND research and care (Worthington 1996, McLeod and Clarke 2007, Pagnini, Simmons et al. 2012). Considering the importance of maintaining QOL to overall treatment goals in MND, there is insufficient research on the psychosocial impacts of MND on quality of life (Epton, Harris et al. 2009) and a lack of direction on providing psychological, psychosocial and spiritual care (Pagnini 2012, Pagnini, Simmons et al. 2012). Below are research findings on some of the key psychosocial factors related to QOL in people with

MND.

### 3.2.2 Hope

Hope and hopelessness are important psychological factors related to QOL in MND. As a result of interviews exploring the meaning of hope in 30 dying people, Kaye Herth defined hope as an inner power directed toward enrichment of being (Herth 1990). Dr. Jerome Groopman defines hope as the ability to see a path to the future (Groopman 2004). People with MND know nothing can be done to change their fate because MND has no cure. As such, a diagnosis of MND threatens hope, forcing families to rework their life goals, dreams and expectations (McLeod and Clarke 2007, Mitsumoto and Rabkin 2007). In research exploring hopelessness in 136 people with MND, 32% of people with MND were reported to feel moderately to severely hopeless (Plahuta, McCulloch et al. 2002). Feeling a loss of control and loss of purpose in life are predictors of hopelessness in people with MND (Plahuta, McCulloch et al. 2002), and hopelessness is positively correlated with suffering and negatively correlated with QOL (Ganzini, Johnston et al. 1999). Like QOL, hopelessness is not correlated with physical function or decline (Plahuta, McCulloch et al. 2002, Fanos, Gelinas et al. 2008). Rather, it is related to social support, spiritual beliefs, the ability to adapt, living in the moment, and self-transcendence (Fanos, Gelinas et al. 2008).

Hopelessness is often considered a component of depression, but people with MND do not suffer from significantly high rates of depression (Neudert, Wasner et al. 2004, Averill, Kasarskis et al. 2007) . A study comparing depression in people with MND and terminal cancer found similar rates of depression in the two groups, although people with MND scored higher on hopelessness and suicidal ideation (Clarke, McLeod et al. 2005). As such, it is the hopelessness facet of depression that appears to differ in people with MND. A 2005 prospective study with 53 participants found that people with MND who displayed hopelessness, loss of meaning, and existential suffering developed a syndrome they termed “end of life despair” and were at increased risk of suicide (Albert 2005 p. 73).

In people with terminal illness, hopelessness is a stronger predictor of suicidal ideation and intent than depression (Chochinov, Wilson et al. 1998). Research shows

that people with MND have an elevated interest in hastened death compared to other disease groups. In Oregon, where physician-assisted suicide (PAS) is legal, 0.4% of cancer deaths were by PAS, while 5% of people with MND chose PAS (Ganzini, Silveira et al. 2002). Those who considered hastening death scored higher on a hopelessness scale than those people with MND who did not (Ganzini, Johnston et al. 1998), and hopelessness is the strongest predictor of assisted suicide in people with MND (Albert 2005). People with MND are also at an increased risk for suicide. A population-based cohort study examining 6,642 people with MND in Sweden between 1965 and 2004 demonstrated a 6-fold increased risk of suicide among persons with MND over the general population. When the study compared people with MND to other terminal illnesses, the MND population had the greatest risk of suicide. Suicide risk was highest in the first year after diagnosis, and higher for people diagnosed at a younger age. These findings are indicative of the psychological distress and hopelessness experienced by people with MND, not only at the end of life but also from the earliest stages of the disease (Fang, Valdimarsdottir et al. 2008).

While depression can be treated pharmacologically, hopelessness requires a non-pharmacological approach to help people with MND see a path to the future, maintain hope, and find meaning in life (Mitchell and Borasio 2007, Simmons 2013). Research exploring hope in people with MND found self-transcendence, a connection to one's past and future generations, reminiscence, relationships, spirituality, and living in the moment led to improved hopefulness (Fanos, Gelinas et al. 2008). Though intervention studies are lacking, psychological interventions addressing these areas are a promising method for decreasing hopelessness among people with MND (Plahuta, McCulloch et al. 2002). It is recommended that psychologists, counselors, spiritual care workers, and other MND health professionals understand hopelessness in order to optimize treatment and psychosocial care (Simmons 2013).

### 3.2.3 Spirituality and meaning

When faced with a life-limiting condition, people often ask questions about the meaning of life (Breitbart, Gibson et al. 2004). Viktor Frankl, one of the founders of existential psychotherapy, stated, "We must never forget that we may also find meaning

in life even when confronted with a hopeless situation, when facing a fate that cannot be changed" (Frankl 2006 at 112). Spirituality has both a religious dimension, relating to a person's relationship with a higher power, and an existential dimension, relating to meaning in life and connection to the world (Dal Bello-Haas, Andrews-Hinders et al. 2000). Psycho-oncology research has shown that spiritual and existential needs are high in cancer patients (Moadel, Morgan et al. 1999). A significant relationship between meaning in life and wellbeing has been reported in people facing death (Fegg, Kogler et al. 2010), and a feeling there is meaning in life appears to safeguard against depression and hopelessness in people with advanced cancer (Breitbart, Rosenfeld et al. 2000). Spirituality offers a source of meaning and comfort as death approaches and, for many, meaning is directly related to spirituality (Fegg, Kramer et al. 2008).

Several studies have documented the importance of religion, faith, spirituality and meaning in preserving QOL in people with MND. A quantitative study with 60 people with MND examining the relationship between spiritual wellbeing and QOL determined that people with higher spirituality had better QOL (Dal Bello-Haas, Andrews-Hinders et al. 2000). In a longitudinal study with 49 people with MND, a significant relationship developed over time between religiousness and improved QOL (Walsh, Bremer et al. 2003). People with MND who possess higher levels of spirituality are more hopeful and have fewer concerns about death (Murphy, Albert et al. 2000). Those who attend worship services and pray regularly report these practices help them to cope with MND (Murphy, Albert et al. 2000). In a qualitative study, people with MND placed the most importance in faith in helping them to accept their illness and see value in their remaining days (Foley, O'Mahony et al. 2007). People with MND who have higher levels of spirituality are less likely to consider assisted suicide (Ganzini, Johnston et al. 1998), and those who find meaning in life despite advancing illness report a strengthened will to live (Ozanne, Graneheim et al. 2013). Religious faith appears to have a protective effect against depression (Rabkin, Wagner et al. 2000), and people with MND describe spirituality as a significant source of strength in coping with the disease and improving QOL (Pagnini, Rossi et al. 2010).

Despite findings such as these, there is no research evaluating the effects of spiritually-based practices or counseling in people with MND (Pagnini, Lunetta et al.

2011). Several interventions, including dignity therapy (see below), have been developed to enhance meaning at the end of life, primarily in cancer patients (Breitbart, Gibson et al. 2004, Chochinov, Kristjanson et al. 2011), but they have not been tested for people with MND.

### 3.2.4 Dignity, social support and relationships

Empirical research on the meaning of dignity to people who are terminally ill has shown that dignity has much to do with social interactions (Chochinov, Hack et al. 2002b). Privacy violations, lack of social support, callous and uncompassionate care, and feeling a burden to others are reported to be perceived as assaults on dignity (Chochinov, Hack et al. 2002a). How a patient sees themselves and is seen by others also affects dignity; the ability to maintain pride, retain autonomy and control, and preserve identity in the face of physical deterioration are important (Chochinov, Hack et al. 2002a). Conserving dignity, where people with MND describe the importance of being respected, valued, and seen independently of their illness, is a common theme in interviews assessing QOL with people with MND (Bolmsjo 2001, Foley, O'Mahony et al. 2007, Foley, Timonen et al. 2012). Protecting self-esteem, maintaining control, and preserving self-image are other important dignity related themes (King, Duke et al. 2009, Ray, Brown et al. 2012). Like hopelessness, loss of dignity is related to wishes for a hastened death in people with terminal illness (Van der Maas, Van Delden et al. 1991, Ganzini, Nelson et al. 2000) and people with MND may request PAS due to dignity related concerns such as feelings of being a burden, loss of autonomy, and loss of dignity (Bascom and Tolle 2002, Mitsumoto and Rabkin 2007).

The importance of good quality family relationships and support affect feelings of dignity and psychosocial health in people with MND (Foley, O'Mahony et al. 2007, Ray, Brown et al. 2012). As a person with MND becomes more physically impaired, the factors which contribute to QOL shift from those that are dependent on physical function, like exercise, hobbies and work, to those that are not, such as social, spiritual and existential factors (Cupp, Simmons et al. 2011). In research on QOL measures, support of family and friends has the strongest impact on the QOL of people with MND (Neudert, Wasner et al. 2004), and Fegg (2010) found people with MND list family,

partners and friends as the greatest sources of meaning in life.

Family carers are among the most important resources for coping in people with MND, and thus problems or conflicts in the relationship are detrimental to QOL (Fegg, Kogler et al. 2010). Feeling a burden on family members, reduced socialization and a perceived lack of social support are frequently cited as psychosocial stressors that affect dignity in people with MND (Ganzini, Johnston et al. 1999, Simmons, Bremer et al. 2000). Moreover, people with MND who have lost the ability to speak encounter higher levels of psychosocial distress due to their social isolation and the impact of the loss on family relationships (Goldstein, Adamson et al. 1998, Ray, Brown et al. 2012). The use of AAC methods can preserve communication between people with MND and their families and social groups and decrease psychosocial distress (Pagnini 2013). Dignity therapy and other interventions that preserve and enhance dignity, self-worth, and accomplishments may be useful to alleviate psychosocial distress in people with MND (Clarke, McLeod et al. 2005)

### 3.2.5 Discussion

The psychological impacts of MND are severe and the impacts on QOL can be significant. Despite progressive physical deterioration and increasing impairment, psychological health and QOL in people with MND is primarily dependent on spiritual, existential, relationship and support factors (Simmons, Bremer et al. 2000, Robbins, Simmons et al. 2001). Researchers examining QOL and psychological distress often conclude that people with MND would experience significant benefits from existential, psychological and psychosocial interventions (Pagnini 2012). Numerous studies mention the need for MND health care providers to consider the psychological, psychosocial and spiritual support systems of people with MND, rather than focus primarily on physical disability (Plahuta, McCulloch et al. 2002, Foley, O'Mahony et al. 2007, McLeod and Clarke 2007). The development of interventions that address hope, meaning, and existential and psychosocial distress in people with MND has often been suggested in the literature (Trail, Nelson et al. 2004, Foley, O'Mahony et al. 2007, Fegg, Kogler et al. 2010). Interventions which engage the family are also important, as research indicates that partner and family relationships have high importance in people

with MND, and family problems or conflict are detrimental to the QOL of people with MND (Fegg, Kogler et al. 2010).

While it is suggested that psychologists have a significant role to play "in every step" of MND (Pagnini, Rossi et al. 2010 p. 1), there is a lack of research and guidance in regard to providing psychosocial and psychological care (McLeod and Clarke 2007, Pagnini 2012) and there are few psychological interventions developed or tested to treat psychosocial and spiritual distress in people with MND (Pagnini 2012). A recent randomized controlled trial of an expressive disclosure intervention, where people with MND wrote or spoke about their deepest thoughts and feelings related to MND over a period of days, found the intervention increased psychological wellbeing in people with MND who previously had difficulty expressing emotions (Averill, Kasarkis et al. 2013). A mindfulness meditation training intervention to reduce distress has been specifically created for people with MND and a randomized controlled trial is currently underway (Pagnini, Di Credico et al. 2014). Nonetheless, people with MND remain largely unaided in finding coping strategies and accessing support to help sustain psychological wellbeing and QOL as the disease progresses. This has resulted in numerous calls to develop and research psychological interventions (Plahuta, McCulloch et al. 2002, Pagnini, Simmons et al. 2012, Whitehead, O'Brien et al. 2012). To guide future intervention research, previous studies have determined that hopelessness, spirituality and meaning, and social support are among the most important psychosocial factors affecting quality of life in people with MND (McLeod and Clarke 2007). Because of its ability to bolster hope, dignity, spirituality and meaning, dignity therapy has been mentioned as a specific intervention that shows promise at alleviating end of life distress in people with MND (Clarke, McLeod et al. 2005, Fanos, Gelinas et al. 2008).

## 3.3 Psychosocial Needs of MND Family Carers

### 3.3.1 Quality of life

Palliative care recognizes that addressing the needs of family carers is a primary objective (World Health Organization 2014). This is especially important for MND family carers, as they provide the majority of care and support to people with MND (Love, Street et al. 2005) , and they play an important role in clinical decision making

(Mitsumoto, Bromberg et al. 2005, Hogden, Greenfield et al. 2013). Research addressing QOL in MND family carers has documented the negative influence of MND on caregiver QOL (Jenkinson, Fitzpatrick et al. 2000, Miyashita, Narita et al. 2011, Aoun, Bentley et al. 2013). While QOL in people with MND remains constant with time, the QOL of MND family carers declines over time (Roach and Averill 2009). QOL in MND family carers appears unrelated to the severity of MND, patient QOL, or the level of care (Rabkin, Wagner et al. 2000, Lo Coco, Lo Coco et al. 2005, Murphy, Felgoise et al. 2009). Factors that contribute to decreased QOL include tension, infrequent outings, decreased time for oneself, and the presence of anxiety and depression (Krivickas, Shockley et al. 1997, Calvo, Moglia et al. 2011, Simmons 2013). Trail and colleagues (2004) found the greatest stressor of MND family carers was existential distress, including loss of faith and worries about the future. Increased QOL in MND family carers is related to religiousness (Calvo, Moglia et al. 2011) and spirituality (Lo Coco, Lo Coco et al. 2005), and MND family carers rate family and social support as one of the most important areas contributing to their QOL (Lo Coco, Lo Coco et al. 2005).

Interventions aimed at psychosocial factors are key to maintaining QOL in MND family carers (Simmons 2013); however, research and development of psychosocial interventions for MND family carers are lacking (Aoun, Bentley et al. 2013). Research findings also suggest the support currently offered to MND family carers by health care providers focuses on practical rather than emotional adjustments (Brown 2003, Oyebode, Smith et al. 2013), which may add to social isolation and inhibit meaning-making processes (Oyebode, Smith et al. 2013). Below are some of the significant psychosocial factors related to QOL in MND family carers.

### 3.3.2 Anxiety and depression

Depression and anxiety are the most common sources of psychological distress in MND family carers (Aoun, Bentley et al. 2013). The rapid deterioration typical in MND results in increased distress over time, including an increase in depression (Goldstein, Atkins et al. 2006, Gauthier, Vignola et al. 2007). Factors related to depression include the ongoing losses that occur as illness progresses (Ray and Street

2006b), the increase in carer burden (Gauthier, Vignola et al. 2007), and the rise in caregiving intensity (Miyashita, Narita et al. 2009). However, a longitudinal study on depression in MND family carers found that psychological distress, rather than clinical depression, is a more accurate description of the depressed mood encountered in MND family carers (Rabkin, Albert et al. 2009). Though some MND family carers in this study had scores on the Beck Depression Inventory-II (Beck, Steer et al. 1996) indicating they were depressed, comments in qualitative interviews suggest that feelings resulting from the caregiving experience, including sadness, feeling under pressure, lacking time for oneself, isolation, and exhaustion, were the cause of depressed mood (Rabkin, Albert et al. 2009).

MND family carers have been found to be more depressed than people with MND receiving care (Trail, Nelson et al. 2003). A study of people with MND/family carer couples found that while QOL and depression levels were steady in people with MND over a nine-month period, MND family carers experienced a significant increase in caregiver burden and depression (Gauthier, Vignola et al. 2007). Caregiver depression may be related to depression in the person with MND (Rabkin, Wagner et al. 2000, Chio, Gauthier et al. 2005), and research suggests that a reason for this is that the consequences of MND influences both the person with MND and their family carer to a similar extent (Olsson Ozanne, Strang et al. 2010). The quality of the relationship between people with MND and their family carers was found to be the most important factor in determining whether an MND family carer developed depression (Rabkin, Albert et al. 2009).

Depression in MND family carers may also be influenced by gender, culture or ethnicity. A study of Japanese MND family carers reported extremely high rates of depression (61%) (Miyashita, Narita et al. 2009), while two studies of American MND family carers reported low rates (19% and 23%), even when patients were severely disabled (Rabkin, Wagner et al. 2000, Rabkin, Albert et al. 2009). There are indications that MND family carers who are women experience higher levels of psychological distress than men though the reasons for this are unclear (Goldstein, Adamson et al. 2000, Hecht, Graesel et al. 2003, Chio, Gauthier et al. 2005, Atkins, Brown et al. 2010, Chio, Vignola et al. 2010).

Only two published studies have assessed anxiety in MND family carers. Goldstein and colleagues (1998) reported 42% of MND family carers had scores suggesting the presence of anxiety, while Vignola (2008) reported prevalence rates from 71 to79%. Risk factors associated with anxiety are longer marriages or shorter duration of symptoms (Goldstein, Adamson et al. 1998). While it has been asserted that anxiety may be associated with the level of disability of the person with MND (Jenkinson, Fitzpatrick et al. 2000, Bolmsjo and Hermeren 2003), or may arise from uncertainty about disease progression (Ray and Street 2006b), these studies did not investigate these links. In qualitative interviews, MND family carers report a wide range of anxieties about the final stage of the disease and death including; fear and uncertainty about the future, worries about losing control and decision making abilities, and anxiety over how they and their children (including adult children) will cope with loss and bereavement (Whitehead, O'Brien et al. 2012). In an Italian study with 40 MND family carers, anxiety, depression and caregiver burden were positively related to each (Pagnini, Rossi et al. 2010).

### 3.3.3 Caregiver burden

For MND family carers, caregiver burden increases over time as physical function of people with MND declines (Goldstein, Atkins et al. 2006, Gauthier, Vignola et al. 2007, Pagnini, Rossi et al. 2010, Aoun, Bentley et al. 2013). MND family carers do not have adequate time for self-care and to attend to their own needs (Rabkin, Albert et al. 2009). They commonly express concerns about their health (Pagnini, Rossi et al. 2010), and report they suffer from exhaustion and fatigue (Rabkin, Wagner et al. 2000, Ray and Street 2006a, Ray and Street 2006b, Gauthier, Vignola et al. 2007, Rabkin, Albert et al. 2009, O'Brien, Whitehead et al. 2011b, O'Brien, Whitehead et al. 2012), as well as insomnia and interrupted sleep (van Teijingen, Friend et al. 2001, Dawson and Kristjanson 2003, Ray and Street 2006a). The lack of time for oneself is reported by some researchers as being a principal reason for caregiver burden (Chio, Gauthier et al. 2005, Gauthier, Vignola et al. 2007, Miyashita, Narita et al. 2009, Pagnini, Rossi et al. 2010).

In a longitudinal study, Gauthier (2007) found burden in MND family carers increased over time, but QOL remained stable. As such, QOL appears dependent on factors other than the physical decline of the care recipient (Lo Coco, Lo Coco et al. 2005). This is identical to the findings on QOL in people with MND, where QOL is not related to physical decline but instead related primarily to psychosocial factors (Simmons, Bremer et al. 2000). However, symptoms of cognitive and neurobehavioral decline (Lillo, Mioshi et al. 2012) and reliance on assisted ventilation both have a significant impact on increasing burden and diminishing QOL in MND family carers (Aoun, Bentley et al. 2013). A recent Australian postal survey of 140 MND family carers identified abnormal behavior in people with MND, including impulsivity and disinhibition, as the strongest predictor of high caregiver burden (Lillo, Mioshi et al. 2012). The increase in caregiver burden and decrease in QOL over time is likely due to increased caring hours, increased anxieties, and decreased social support that can result from cognitive and behavioral impairment, constant mechanical ventilation, or other factors (Aoun, Bentley et al. 2013) rather than physical decline in and of itself.

Positive social support mitigates burden in MND family carers (Lo Coco, Lo Coco et al. 2005, Love, Street et al. 2005, O'Connor, McCabe et al. 2008, Atkins, Brown et al. 2010, Pagnini, Rossi et al. 2010). Satisfaction with social support levels weakens the negative effects of MND on perceived burden and QOL (O'Connor, McCabe et al. 2008, Atkins, Brown et al. 2010). However, MND family carers, particularly those providing care over an extended time, encounter difficulties seeking and maintaining social support (Chio, Gauthier et al. 2005, Love, Street et al. 2005, Ray and Street 2006a). MND family carers tend to have reduced social contacts, decreased social networks and poor quality social interactions due to caregiving responsibilities (Trail, Nelson et al. 2003, Love, Street et al. 2005, Ray and Street 2005), which contribute to social isolation and perceived burden (Love, Street et al. 2005). When social supports are reduced, MND family carers have an increased reliance on professional services; however, research has shown that adequate professional supports are lacking and MND family carers are dissatisfied with the lack of available services (O'Brien, Whitehead et al. 2012) which indicates a gap in care.

### 3.3.4 Hope

Few studies have evaluated the impact of hope and hopelessness on family carers in a palliative care setting (Williams, Duggleby et al. 2013). However, in the studies that have been conducted, hope has been identified as a psychosocial resource that helps family carers of the terminally ill to cope with the difficulty of the caregiving experience (Herth 1993, Dawson and Kristjanson 2003, Duggleby, Holtslander et al. 2010). Hope impacts on a family carer's ability to cope with stressful, life-threatening situations (McClement and Chochinov 2008) and higher hope levels are associated with family carers feeling prepared to take on the tasks and demands of the caregiver role (Henriksson and Arestedt 2013).

Family carers of the terminally ill define hope as "a dynamic inner power that enables transcendence of the present situation and fosters a positive new awareness" (Herth 1993 p. 538). In a meta-synthesis of 14 qualitative studies on hope in family carers of people with chronic conditions, Duggleby and colleagues (2010) defined hope as "transitional dynamic possibilities within uncertainty" by conceptually integrating the synthesized themes found in the studies (p. 154). In a longitudinal study with 25 family carers of the terminally ill where interviews occurred at three time points, from two weeks after hospice admission to two weeks before death, hope changed over time from specific hopes to more general hopes, such as hope for a close relationship with the family member or for a peaceful death (Herth 1993). Findings from a recent study of hope in caregivers of advanced cancer patients found hope was related to social support, spirituality, living in the moment, acceptance, and optimism (Williams, Duggleby et al. 2013). Hope is important to family carers regardless of age, relationship or setting (Duggleby, Holtslander et al. 2010), although younger family carers are at risk for both lower levels of hope and higher levels of distress (Lohne, Miaskowski et al. 2012).

In contrast, in qualitative interviews with 9 bereaved family carers in Finland, a lack of hope, where there is little to look forward to in the remaining days of their family member, was defined as a feature of poor palliative care (Miettinin, Alaviuhkola et al. 2001). In a study of 10 Canadian family carers of people receiving palliative care, "eroding hope" was a main concern, which resulted from bad days, negative messages,

and poor experiences in the health care system (Holtslander, Duggleby et al. 2005). Family carers dealt with eroding hope by living in the moment, acceptance, staying positive, and maintaining a fighting spirit (Holtslander, Duggleby et al. 2005). Studies have shown family carers of the terminally ill score lower levels of hope overall than the patients themselves (Herth 1993, Benzein and Berg 2005). Increasing hope may help decrease depression and caregiver distress (Lohne, Miaskowski et al. 2012). To increase hope in family carers of advanced cancer patients, a journaling program, focusing on what gives a carer hope each day, are currently being developed and tested (Williams, Duggleby et al. 2013).

While studies have evaluated the influence of hope in family carers of the terminally ill, none have studied its impact on MND family carers. However, studies with MND family carers have highlighted hopelessness as an important factor related to psychological distress. MND family carers said having something to look forward to was crucial to their ability to go on (Rabkin, Albert et al. 2009). In a qualitative study with 16 bereaved MND family carers, 11 family carers, including all 6 who met the criteria for prolonged grief disorder, expressed profound feelings of hopelessness during their caregiving experience (Aoun, Connors et al. 2012). Research by Chio and colleagues (2005) concluded that caregiver burden may be alleviated in MND family carers by supporting a sense of hope. These findings suggest that interventions that assist family carers to increase hope and decrease hopelessness may help to reduce depression and alleviate distress.

### 3.3.5 Discussion

Research has shown a high degree concordance between psychological distress in MND family carers and the psychological distress of people with MND, specifically in the areas of depression, anxiety and quality of life (Rabkin, Wagner et al. 2000, Chio, Gauthier et al. 2005). The QOL, distress and burden experienced by MND family carers appears to be directly related to the existential wellbeing and spirituality of the care recipient (Pagnini, Lunetta et al. 2011). In a longitudinal study where people with MND and their family carers rated the wellbeing of both themselves and their counterpart, the psychological wellbeing of people with MND and their family carers retained a close

correlation over time that was not affected by disease progression or increased disability (Olsson, Markhede et al. 2010). Similarly, Atkins et al (2010) studied marital relationships in MND and found the quality of the marital relationship was related to psychological and emotional factors rather than symptoms of the disease. Findings from palliative care research shows that hope within family members has a reciprocal role in maintaining hope in terminally ill patients (Herth 1990, Herth 1993). These findings indicate the importance of treating psychosocial distress in MND family carers as it may also positively affect the person with MND, and also in the reverse (Rabkin, Wagner et al. 2000).

While research continues to document the burden and distress experienced by MND family carers and the concordance of distress between people with MND and their family carers, this evidence has not translated into improved care through access to psychological or psychosocial supports (Aoun, Bentley et al. 2013). Interviews with MND family carers have found that they desire psychological and psychosocial support, but are often unable to access these services (O'Brien, Whitehead et al. 2012). This disparity between desired supports and the availability of supports is a source of carer distress (O'Brien, Whitehead et al. 2012). These studies highlight a gap in care, and there are no intervention studies to date that have developed or assessed direct psychosocial supports for MND family carers (Aoun, Bentley et al. 2013) despite suggestions in the literature (Benzein and Berg 2005, Pagnini 2012). However, recommendations for future interventions are suggested in many of the studies.

Interventions designed to enhance communication between people with MND and their family carers have the potential to improve the psychological wellbeing of both parties (Gauthier, Vignola et al. 2007). Counseling interventions, care information courses and support groups have the potential to reduce the burden of MND family carers (Hecht, Graesel et al. 2003). The development of spiritually-based interventions have been indicated by several studies to potentially improve MND family carers' QOL (Davey, Wiles et al. 2004, Trail, Nelson et al. 2004, Lo Coco, Lo Coco et al. 2005, Murphy, Felgoise et al. 2009, Calvo, Moglia et al. 2011). Interventions that improve existential wellbeing in people with MND may also improve the psychological wellbeing of their family carers (Pagnini, Rossi et al. 2010). Dignity therapy

(Chochinov, Hack et al. 2005), which focuses on maintaining hope and a sense of meaning in the patient, may also increase hope and meaning in the family carer. Caregiver burden may be decreased by an intervention like dignity therapy, as burden is decreased in those who find meaning in their caregiving experience (Chio, Gauthier et al. 2005).

## 3.4 Dignity therapy

### 3.4.1 History and empirical foundation

Professor Harvey Max Chochinov is a Canadian psychiatrist and researcher working in the field of palliative care. Influenced by Dame Cicely Saunders' concept of total pain (Chochinov 2012), he has become a strong advocate for person-centered care at the end of life. In the 1990s, Chochinov and colleagues studied why some people with terminal illness sought a hastened death and found a strong association between a desire for hastened death and hopelessness and depression (Chochinov, Wilson et al. 1995, Chochinov, Wilson et al. 1998). Loss of dignity was a common source of distress for those with terminal illness and was identified as a primary reason why patients lost the will to live (Van der Maas, Van Delden et al. 1991, Ganzini, Nelson et al. 2000, Chochinov, Hack et al. 2002b), but "dignity" was a vague term subject to multiple meanings (Chochinov, Hack et al. 2002a). Chochinov then sought to understand the meaning of dignity from the perspective of a dying person through qualitative interviews with 50 terminally ill patients. Through this research, Chochinov developed an empirical model of dignity in the terminally ill where he identified three major categories with themes and subthemes, shown in Table 3-1 (Chochinov, Hack et al. 2002a). Chochinov validated his dignity model in a subsequent quantitative study where 211 advanced cancer patients were asked to rate the extent to which their personal dignity was related to the 22 individual items derived from the themes and subthemes in Figure 3-1. In this study, all but 1 of the 22 items (thinking about how life would end) were endorsed by over half of the participants (Chochinov, Kristjanson et al. 2006).

**Table 3-1. Major dignity categories, themes and subthemes, with example statements in italics**

| Illness Related Concerns | Dignity Conserving Repertoire | Social Dignity Inventory |
|---|---|---|
| Level of Independence<br>Cognitive Acuity<br>*"Can I think clearly?"*<br><br>Functional Capacity<br>*"Can I function physically?"*<br><br>Symptom Distress<br>Physical Distress<br>*"Am I experiencing physical pain?"*<br><br>Psychological distress<br>*"What will happen with my medical condition?"*<br>*"How will I die?"* | Dignity Conserving Perspectives<br>Continuity of self<br>*"Am I still the same person?"*<br>Role Preservation<br>*"Can I still play important roles?"*<br>Generativity/legacy<br>*"Am I leaving something behind?*<br>Maintenance of pride<br>*"Can I still take pride in myself?*<br>Hopefulness<br>*"Do I still have hope?"*<br>Autonomy/control<br>*"Do I feel in control?"*<br>Acceptance<br>*"Am I at peace with what is happening?"*<br>Resilience/fighting spirit<br>*"Do I have the will to go on?"*<br>Dignity Conserving Practices<br>Living "in the moment"<br>*"Am I able to live each day as it comes?*<br>Maintaining normalcy<br>*"Am I able to have some normality to my days?*<br>Seeking spiritual comfort<br>*"Can I find comfort in my spiritual beliefs?"* | Privacy boundaries<br>*"Are my privacy boundaries being violated?"*<br><br>Social support<br>*"Am I supported by my family and friends?"*<br><br>Care tenor<br>*"Am I being treated by my health professionals with respect and kindness?*<br><br>Burden to others<br>*"Am I a burden to others?"*<br><br>Aftermath concerns<br>*"What will happen to those I love after I die?"* |

Chochinov used his empirical model of dignity in the terminally ill to pursue research in two directions: 1) the development of "dignity-conserving care," a model for palliative care providers designed to improve the health care experience by preserving the dignity of those nearing death (Chochinov, Hack et al. 2002a, Chochinov 2007); and 2) the development of dignity therapy, a brief psychotherapeutic intervention to bolster dignity and alleviate distress at the end of life (Chochinov, Hack et al. 2005). Through

his research, Chochinov found that as a person moves closer to death, feelings of hopelessness, being a burden to others, and a loss of dignity are important psychological stressors that often lead to existential suffering and depression (Chochinov 2006). As a psychiatrist, Chochinov sought to explore the use of psychotherapy to alleviate these stressors. Before developing dignity therapy, Chochinov and colleagues evaluated other psychotherapeutic interventions already in use, including group therapy, cognitive behavioral therapy, logotherapy, and life review. They concluded that none of these were ideal as they did not specifically address the types of dignity-related issues that confront the dying (Chochinov, Hack et al. 2004). Moreover, they found psychotherapy was not widely embraced in palliative medicine, and there was a lack of research and efficacy studies supporting the use of different modalities in palliative care populations (Chochinov, Hack et al. 2004).

Chochinov's empirically based model of dignity at the end of life provided the framework for the development of dignity therapy and informs the tone, structure and content of the therapy. Dignity therapy touches on seven specific dignity-related themes from Chochinov's empirical model of dignity, shown in Figure 3-1 (Chochinov, Hack et al. 2005). How dignity therapy specifically addresses the seven dignity concerns is shown in Table 3-2 (Chochinov, Hack et al. 2005).

**Figure 3-1. Dignity concerns supported by dignity therapy**

**Table 3-2. Dignity concerns and how dignity therapy addresses the concern**

| DIGNITY CONCERN | HOW DIGNITY THERAPY ADDRESSES THE CONCERN |
|---|---|
| **Generativity** | Interview sessions between the therapist and client are tape-recorded and transcribed, with an edited transcript returned to the client who may share it with and bequeath it to family and/or friends. |
| **Continuity of self** | Clients are invited to share memories that are foundational to their identity and sense of self. |
| **Role preservation** | Clients are invited to share previous or current roles that contribute to their core self. |
| **Maintenance of pride** | Clients have the opportunity to speak about their accomplishments and other sources of pride. |
| **Hopefulness** | Clients are engaged in a therapeutic process intended to increase hope through discovery meaning and purpose. |
| **Aftermath concerns** | Clients are given the opportunity to help prepare their family for a future without them. |
| **Care tenor** | A dignity therapist is empathic, nonjudgmental, encouraging and respectful, and through the therapy experience sees the client as a whole person rather than as a dying patient. |

### 3.4.2 The intervention

Dignity Therapy was designed to decrease suffering, enhance quality of life, create spiritual peace, strengthen relationships, and increase a sense of meaning, purpose and dignity at the end of life (Chochinov, Hack et al. 2005). In dignity therapy, a therapist invites a person with life-limiting illness to discuss aspects of their life they want remembered and share important messages using the dignity therapy question protocol (Chochinov, Hack et al. 2005, Chochinov 2012). The protocol is composed of several questions as follows:

- Tell me a little about your life history; particularly the parts that you either remember most or think are the most important? When did you feel most alive?
- Are there specific things that you would want your family to know about you, and are there particular things you would want them to remember?

- What are the most important roles you have played in life (family roles, vocational roles, community service roles, etc.)? Why were they so important to you, and what do you think you accomplished in those roles?
- What are your most important accomplishments, and what do you feel most proud of?
- Are there particular things you feel still need to be said to your loved ones or things that you would want to take time to say once again?
- What are your hopes and dreams for your loved ones?
- What have you learned about life that you would want to pass along to others? What advice or words of guidance would you wish to pass along to your son, daughter, husband, wife, parents, other(s)?
- Are there words or instructions that you would like to offer your family to help prepare them for the future?
- In creating this permanent record are there other thing you would like included?

The sessions are recorded, transcribed and carefully edited together with the participant. A final document, which Chochinov calls a 'generativity document' (Chochinov, Hack et al. 2005, Chochinov 2012), is returned to the participant, which they can share or bequeath to family members and friends. In using the term 'generativity," Chochinov borrows from Erik Erikson's theory of psychosocial development (Chochinov and McKeen 2011) where Erikson described 8 developmental tasks appropriate to all individuals, spanning from birth to death (Erikson 1950). In the seventh stage, the task of 'generativity' encompasses giving back to society and helping to guide future generations. Thus, the 'generativity document' resulting from dignity therapy is a means to document a person's legacy for future generations. Below is a step-by-step summary of the dignity therapy process (Chochinov and McKeen 2011, Chochinov 2012):

Initial meeting: In the first session, the therapist explains how dignity therapy works, introduces the dignity therapy questions, identifies potential recipients of the generativity document, and answers any questions. At the conclusion of this meeting, the time and setting for the interview are discussed and arranged.

The interview: A guided recorded conversation takes place between the therapist and participant using the dignity therapy questions in order to address important issues

the participant would like to document, such as personal history, meaningful roles and accomplishments, lessons learned, and hopes or wishes for their family.

Editing: A verbatim transcript of the recorded interview is created. The therapist who performed the interview performs an initial edit of the transcript outside the presence of the participant changing the verbatim interview into a more prose-like document. The purpose of this initial editing process is to clean up the transcript, clarify what is said, correct time sequences and find a suitable ending. After the manuscript has been shaped and polished, another meeting takes place where the therapist reads the document to the participant. Together they make any final edits to the document. The participant may choose to add or delete material until they feel the document reflects what they truly want to say.

Finalizing the document and concluding the therapy: The therapist finalizes the document and presents copies to the participant, who can share them as they wish. The document is printed and bound in a way that will preserve the document and which reflects its importance. For example, Chochinov's practice is to print the document on "bond grade, beige paper, and to place it in blue, lightweight binder" (Chochinov 2012 p. 98)

Dignity therapy invites participants to talk about the parts of their life that they found most meaningful. It also allows them to talk about issues related to grief, offer comfort to their loved ones, and provide advice and instructions for the future. The process creates a tangible legacy, the dignity therapy generativity document, and provides the opportunity for the participant to leave behind a permanent record of his or her life (Chochinov, Hack et al. 2005, Chochinov 2006).

### 3.4.3 Previous research

Dignity therapy was pilot tested by Chochinov and colleagues with 100 participants, 97 of whom had end-stage cancer. This one group, pre/post-test study found that dignity therapy was acceptable to participants, who reported numerous benefits from the therapy, and it showed a promising ability to alleviate distress at the end of life (Chochinov, Hack et al. 2005). Sixty family members completed a feedback questionnaire several months after the death of the participant. Ninety-five percent of

family members reported that dignity therapy had helped the deceased before death, and 78% reported that the document helped them in bereavement (McClement, Chochinov et al. 2007). Thus, dignity therapy showed a promising potential to reduce distress in family members of the terminally ill, as well as to mitigate their bereavement experience.

Subsequently, the National Institutes of Health funded an IRCT. In that study, 326 participants (95% of whom had cancer) were randomized into three arms – dignity therapy, client-centered care, and standard palliative care. There were no significant differences reported on outcome measures of distress between the three groups post-intervention. However, the study demonstrated greater self-reported benefits in the dignity therapy group similar to those found in the pilot study. As a result, dignity therapy was recommended for clinical use with patients nearing death (Chochinov, Kristjanson et al. 2011).

While it may be considered somewhat unusual for a psychotherapy that did not demonstrate significant differences on the outcome measures to be recommended for clinical use, Chochinov made a convincing case for why his study failed to show improvements on the outcomes but was still beneficial to patients (Chochinov, Kristjanson et al. 2011). Specifically, the study population reported low baseline levels of distress prior to the intervention and therefore floor effects prevented improvements in the outcomes (Chochinov, Kristjanson et al. 2011). Chochinov cited a 2010 meta-analysis which concluded that distress prior to an intervention is critical when researching psychosocial interventions in people with cancer (Schneider, Moyer et al. 2010). Chochinov also pointed to a recent published review paper which found there are no quality of life measures responsive to change in palliative care populations (Albers, Echteld et al. 2010). Finally, while palliative care is intended to address the spiritual and psychosocial needs of patients and their total pain, Chochinov noted there are extremely few psychotherapeutic options for use at the end of life to address these concerns. Consequently, he argued that dignity therapy's 'something,' which are the numerous benefits reported by study participants facing death, is better than nothing, and that the findings were sufficient to support dignity therapy's continued clinical use (Chochinov, Kristjanson et al. 2011). In an environment where it is recognized that palliative care

research is exceptionally challenging (McWhinney, Bass et al. 1994, Grande and Todd 2000), Chochinov's argument was embraced and dignity therapy was widely endorsed (Ferrell 2005, Nekolaichuk 2011, Akechi 2012, Hack 2012, Kumar, Morse et al. 2012). Soon after publication of the results of the IRCT, Chochinov published an instructional manual for the clinical application of dignity therapy (Chochinov 2012) and routine clinical practice of dignity therapy was initiated in hospice environments in several countries, including the U.S. and Australia (Squires 2014, Zahn 2014).

### 3.4.4 Recent developments

Twenty-three additional studies have been published in recent years evaluating various aspects of dignity therapy, from its use with different cultural groups, different disease populations, the cost of implementing the therapy, the experiences and perspectives of staff delivering the therapy, and the themes found in dignity therapy documents. This research is summarized below and in Appendix A.

#### 3.4.4.1 Feasibility studies with different study populations

Two studies evaluated the feasibility of providing dignity therapy to elderly people in residential care, expanding dignity therapy from a palliative care intervention used in the final months of life to general use with people receiving care in the final years of life. Similar to the results of the IRCT, a phase II randomized controlled feasibility study of dignity therapy performed with 60 older people in care homes in the UK found no significant differences in the outcome measures, but the dignity therapy group reported more benefits than the control group in a feedback questionnaire. The results suggest dignity therapy may be beneficial to this non-palliative care population, though it may take longer to perform (Hall, Goddard et al. 2012). In a second feasibility study with 23 elderly people in long-term care in Canada, dignity therapy was tested with two groups: 1) those who were cognitively intact and who completed the therapy themselves, and 2) those who suffered from dementia and completed the therapy through a family member proxy. In this study, the participants, proxies, and health care providers reported benefits in a feedback questionnaire. The findings suggest that dignity therapy can be successfully performed through a proxy with people who have cognitive impairment which is a substantial modification of the original intervention,

and demonstrated that it may provide benefits to the elderly in aged care settings. The authors also suggest that the dignity therapy document can assist health care providers in aged care settings to treat all residents with dignity, thus improving care (Chochinov, Cann et al. 2012).

A feasibility study of dignity therapy delivered to 8 people using videoconferencing was undertaken to evaluate whether dignity therapy could be delivered to people dying at home, including those in rural and remote locations. The small study, conducted over a decade ago when videoconferencing from home was less common, found that telemedicine appeared to be a feasible way to deliver dignity therapy. The intervention was delivered in a timely fashion with few technical issues, and participants reported high satisfaction with dignity therapy in a feedback survey. This study noted, however, that therapists commonly complained about the amount of time needed to edit the transcripts into a final document (Passik, Kirsh et al. 2004).

#### 3.4.4.2 Effectiveness of dignity therapy

Several studies have examined the effectiveness of dignity therapy to validate and expand on the findings of the dignity therapy IRCT. A Danish study found similar benefits were reported on a feedback survey as were reported in the IRCT. The pre/post-test evaluation performed a culturally modified form of dignity therapy with 80 people with advanced cancer who were admitted to inpatient and outpatient palliative care. In addition to the feedback survey, they measured six outcomes at the conclusion of the intervention with mixed results: QOL decreased and depression increased while the participants' sense of dignity and sense of being a burden improved (Houmann, Chochinov et al. 2014). The authors also reported that participants with children appeared to experience an increased benefit, which supports dignity therapy's ability to address generativity concerns and the legacy they are leaving to future generations.

In a qualitative study, interviews were performed with people with advanced cancer in both the intervention and control group with the aim of critically examining the perceptions and benefits encountered during participation in dignity therapy research. Interviews were completed with those who remained in a dignity therapy study at one week ($N$=29) and four weeks ($N$=20) after the document was completed.

Participants from both the intervention and control groups indicated that they received a benefit from their contact with the researchers. There was also support for four of the seven dignity concerns from the model of dignity in both groups, including 'continuity of self,' 'maintenance of pride,' 'care tenor,' and 'hopefulness.' The only difference between the groups was the dignity therapy group supported the concept of 'generativity' concerns. Moreover, the researchers did not find support for 'role preservation' and 'aftermath concerns' in either group. The findings of this study suggest that advanced cancer patients find similar benefits through participating in research as they do from dignity therapy, although dignity therapy may offer an additional benefit by addressing generativity concerns (Hall, Goddard et al. 2013b). However, the study did not find support for two of Chochinov's seven dignity concerns from the 22-item dignity model which form the theoretical basis for dignity therapy.

In order to examine whether social or cultural factors affected the outcomes of dignity therapy, a small phase II randomized controlled trial of dignity therapy with 45 advanced cancer patients was performed in the UK. The results showed improvement on one outcome, hope, which was higher in the intervention group receiving dignity therapy (though hopefulness was not assessed in the IRCT). The intervention group also reported more benefits in a feedback survey than the control group, which is consistent with the findings of the IRCT (Hall, Goddard et al. 2011). As a result of this study, hopefulness emerged as an important outcome measure in dignity therapy research. A second study soon followed which also found improvements in hope through dignity therapy: an RCT in Iran with 70 people with chronic renal failure found that the hope levels in the dignity therapy group increased significantly one month post-therapy. This research also suggests that dignity therapy may be beneficial to people with chronic illness (Vaghee, Javadi et al. 2012).

To address the hypothesis put forth in the findings of the IRCT that floor effects resulting from low baseline levels of distress prevented improvements on the outcome measures, a Portuguese RCT sought to limit the study population to terminally ill patients with high baseline levels of anxiety and depression. Preliminary results from this study with 60 participants, 95% of whom had cancer, found that dignity therapy reduces anxiety and depression at 4 and 15 days post-intervention, although these effects

were not sustained at 30 days post-intervention (Juliao, Oliveira et al. 2012). This research suggests that dignity therapy may be effective in the short-term in reducing anxiety and depression in patients with elevated anxiety and depression scores. On the other hand, a small study with 10 participants with advanced cancer found distressed participants were more likely to drop out and not complete the therapy, indicating that dignity therapy may not be ideal for distressed populations. In a feedback questionnaire, three of the four participants who completed dignity therapy reported that dignity therapy had been helpful and made their life more meaningful (Johns 2013), which is consistent with the findings of the IRCT and other dignity therapy research.

#### 3.4.4.3 Effectiveness of dignity therapy in aged care settings

Following the two studies which found dignity therapy to be feasible for the elderly in residential care, qualitative research was conducted to explore the benefits experienced by this group as a result of dignity therapy. Interviews were conducted with both the control and intervention groups. Both groups indicated they received a benefit by taking part in the study and through interactions with researchers. Interviews revealed that aged care residents gave little thought to the dignity concerns of 'generativity,' 'aftermath concerns,' 'role preservation,' and 'continuity of self.' This study also reported that the benefits of dignity therapy were countered by problems and concerns, as some aged care participants felt the document was not a thorough enough biography and others were concerned how their documents would be received. Though all participants passed a brief cognitive screen, the Blessed Orientation Memory Concentration test (Katzman, Brown et al. 1983), the researchers found cognitive impairment was a factor with minor and major memory lapses affecting the therapy and resultant documents. The findings of this study suggest that dignity therapy may be beneficial to a small minority of aged care residents, but that it should not be offered as usual care (Hall, Goddard et al. 2013a, Hall, Goddard et al. 2013b).

#### 3.4.4.4 The impact of dignity therapy on families

To explore further the results reported in the pilot study where dignity therapy showed a benefit to family members during bereavement (McClement, Chochinov et al. 2007), three studies looked at the impact of dignity therapy on family members. In one

study, interviews were conducted with 14 family members of older people in care homes in the UK in order to explore the family members' views and experiences of dignity therapy performed with their relative. The findings were primarily positive and family members reported the documents helped them learn new details about their relatives, communication with their relatives was enhanced as a result of reading the documents, and many felt the documents had the potential to provide comfort during bereavement. On the other hand, some family members also voiced worries and concerns over the content of the documents and the strain placed on their relatives as a result of the therapy process (Goddard, Speck et al. 2013). In a second study, six family members of advanced cancer patients in the US who received dignity therapy completed a feedback survey. All reported that reading the transcript was meaningful to them and that it increased the sense of purpose, dignity and meaning for their relative (Johns 2013). However, in a third qualitative study with nine family members of advanced cancer patients, two-thirds described negative experiences as a result of their relative's participation in dignity therapy, including concerns about the content of the generativity document, worries about who received and did not receive the document, and fears over its impact on family relationships, although family members remained positive about the overall impact of dignity therapy on the participant, themselves and other family members (Hall, Goddard et al. 2013b). The findings of these studies indicate that family members primarily experience dignity therapy as positive, but the potential exists to create problems and disturbances for family members through the creation of the dignity therapy generativity document.

#### 3.4.4.5 Dignity therapy with different cultural groups

The creator of dignity therapy and the theoretical model of dignity at the end of life upon which dignity therapy is grounded, Professor Harvey Max Chochinov, is based in Canada, and the pilot study and IRCT took place at three international sites: Winnipeg, Canada, New York City, USA, and Perth, Australia (Chochinov, Hack et al. 2005, Chochinov, Kristjanson et al. 2011). As such, dignity therapy is a distinctly Western intervention. Two studies have examined whether dignity therapy can be performed with different cultural groups. A study in Denmark assessed whether the dignity therapy interview questions were acceptable to the Danish cultural group. This

study found dignity therapy was acceptable with some revisions to the question protocol, for example, reducing emphasis on self-praise, pride in oneself and accomplishments, as these were not culturally accepted. The results of this study suggest that dignity therapists must be culturally sensitive and modifications to the question protocol may be necessary for different cultural groups (Houmann, Rydahl-Hansen et al. 2010).

A study in Japan was abandoned when they were unable to recruit eligible advanced cancer patients with less than six months to live to receive dignity therapy and received an 86% refusal rate. The reasons given for refusal suggest cultural differences between Western and Japanese attitudes about the end of life. Aversion to death awareness is more common in Japan and dignity therapy was found to be too confronting for many (Akechi, Akazawa et al. 2012). In a second Japanese study, only subjects expected to benefit from dignity therapy were approached. Eleven patients were recruited and completed dignity therapy. The findings from feedback questionnaires reported satisfaction figures that were not as high as those found in Western studies. As a result, the authors suggest that dignity therapy should not be offered routinely in Japan due to cultural differences, but it may benefit some Japanese patients who wish to leave a legacy (Akechi, Akazawa et al. 2012). These finding suggest that research surrounding the acceptability of dignity therapy be conducted before dignity therapy is adopted for use with non-Western cultural groups.

#### 3.4.4.6 Case studies

Case studies have the ability to provide detail, explore a new area, and develop theoretical insights (Walshe, Caress et al. 2004). Several authors have published case studies to provide depth and pose new questions for future dignity therapy research. A paper that presented case studies of three distressed people with advanced cancer who completed dignity therapy found these participants felt dignity therapy helped them and would help their families at 1 and 4 weeks post-intervention. However, the cases highlighted the complexities of delivering a psychotherapy to distressed people with advanced illness who are struggling with issues dignity therapy is not designed to address, such as physical pain or social problems. In addition to experiencing difficulty

delivering the therapy in these cases, the dignity related problems of these participants returned when their condition deteriorated (Hall, Goddard et al. 2013c).

A New York psychiatrist presented two novel cases of dignity therapy with people suffering from mental illness. In the first, a woman with severe depression who struggled with feelings of hopelessness and loss of meaning was given dignity therapy. The subject reported that it helped her "find hope" and improved her mood (Avery and Baez 2012). In the second case, a man with severe mental illness awaiting transfer to a state hospital for psychiatric hospitalization underwent a modified dignity therapy process where he wrote out answers to the dignity therapy questions and then the psychiatrist created a generativity document from the writings. The participant reported dignity therapy "restored hope" and helped him communicate with family members. Also, the participant's children stated that they understood their father better as a result of dignity therapy (Avery and Savitz 2011). In both studies, the authors argue that people with chronic or severe mental illness have much in common with people who are facing death, including loss of dignity, hopelessness, loss of purpose and meaning, and low QOL. They suggest future research to adapt dignity therapy to be used with people who have a major mental illness (Avery and Savitz 2011, Avery and Baez 2012).

#### 3.4.4.7 Themes in dignity therapy documents

To understand dignity therapy better, researchers have inquired into the content of generativity documents, and four studies examined the themes found in dignity therapy transcripts. Fifty documents from the IRCT were analyzed to discover the most common 'values' expressed in dignity therapy; these were family, pleasure, caring, sense of accomplishment, true friendship, and rich experience (Hack, McClement et al. 2010). In a study where 23 dignity therapy transcripts were examined, the final transcripts were qualitatively analyzed to discover the most commonly discussed areas. In rank order, they were autobiographical information, love, lessons learned, important roles, accomplishments, character traits, unfinished business, hopes, catalysts, overcoming challenges, and guidance for others (Montross, Winters et al. 2011). In both studies, the values and themes expressed by participants bear a relation to the questions posed in the dignity therapy question protocol.

Twelve final dignity therapy transcripts were qualitatively analyzed to examine the narrative types and themes. The study found patients' stories fell into three narrative types: 1) evaluation narratives where patient's reflected on their early life before illness occurred; 2) transition narratives, where patients discussed the transition from the past to their current condition; and 3) legacy narratives, where patients reflected on stories and messages they wished to leave their loved ones. An overarching theme of overcoming adversity was discovered in evaluation narratives, and common subthemes included heritage, maturation and social lives. In transition narratives, themes included illness-related concerns and personal growth through the illness experience. In legacy narratives, themes included newfound perspectives as a result of facing death, messages to family to appreciate life before it was too late, and advice on how family should move forward with their lives (Tait, Schryer et al. 2011). In a second paper analyzing the same transcripts, the authors found participants used 'eulogistic strategies,' to create discursive order out of their life events, moving readers from the past, through the present, and to a future where they are deceased (Schryer, McDougall et al. 2012).

#### 3.4.4.8 Implementation studies

Three studies have sought to provide information about the pragmatic aspects of delivering dignity therapy in a clinical setting, including referral processes, transcription costs, and clinical time required for service provision. In one study in a hospice setting performed in the US, 20 people completed dignity therapy. The mean number of sessions was 4 and the mean total clinical session time per participant was 6.3 hours. (This does not include the editing time which was done solely by the clinician and which was not tracked). On average, patients addressed five questions from the dignity therapy question protocol. The mean finished document was 8 pages in length and 2,993 words long, the cost of transcription ranged from $27 to $144 with a mean of $56 per transcript (Montross, Winters et al. 2011).

A second study was performed examining the implementation of dignity therapy into clinical practice at a cancer center in the US. Ten people entered the study, three declined to complete the editing process and three died before the documents could be completed. Of the four that were finished, the mean document was 11 pages in length

and 5,878 words long, which, for reasons unknown, is almost double the length of those in the Montross study cited above. Therapists spent 3 to 4 hours editing the document and the average interview lasted 66 minutes. There was no information provided on total sessions, total therapist time involved, or duration of the therapy (Johns 2013).

A third study with 27 people in an aged care setting reported the mean total hours taken to deliver the therapy was 15.04 hours per participant. The mean hours of client contact time amounted to 3.48 hours (just over half of the 6.3 hours reported client contact time in the Montross study cited above). The mean hours needed to edit the documents was 8.69 (more than double the 3 to 4 hours reported in the Johns study above). The authors report that the increased editing time was due to disjointed stories and memory issues of the participants, which may also account for the reduced client contact time. The mean duration of the therapy was 31.81 days from initial interview to handing over the document. This study did not report on the length of the document (Hall, Goddard et al. 2012). From the research conducted thus far, it is difficult to assess the time and resources required to implement a dignity therapy program into practice. None of the studies provided comprehensive data and there are major differences across the data presented.

#### 3.4.4.9 Clinical perspectives

Four studies explored the impacts of dignity therapy on staff and clinicians. Feedback was sought from 18 hospice staff members who referred people for dignity therapy on the value of dignity therapy as a clinical service. Overall, staff felt dignity therapy was worthwhile, helpful, had some ability to reduce pain and suffering, and that it would help family members. Staff reported that dignity therapy was a positive experience for patients and also helped increase their connections to patients (Montross, Meier et al. 2013). Similar findings emerged from a survey of 7 health care providers in an aged care setting (Chochinov, Cann et al. 2012).

Thirty-six undergraduate students in a social work class performed dignity therapy with hospice patients and were interviewed about the impact that administering dignity therapy had on them. Three themes emerged; “greater appreciation for life,” “connection to my own family,” and “service and legacy.” The study found that the

experience was a resounding success and a meaningful experience (Davis Berman 2014). In another study, twelve medical residents performed dignity therapy in a study that aimed to examine residents' experiences of delivering dignity therapy, residents' reflections on patients' life stories, and residents' reflections on the dignity therapy intervention in the context of their broader medical education. The study found that residents felt conversations with dying patients, particularly the skill of soliciting the patients' life story, was poorly taught in medical school, and that a hidden message existed in their training that learning a patient's story is not a valued domain of the physician (Tait and Hodges 2013). It is noteworthy that in all of these studies, clinicians reported a benefit to themselves from dignity therapy, often as both a personally meaningful experience for themselves, as well as a method of enhancing care.

### 3.4.5 Discussion

While there were no differences on the outcomes measuring distress between the three study arms in the dignity therapy IRCT, the dignity therapy group reported more benefits and higher satisfaction with the therapy on a feedback questionnaire than the other groups. Therefore, the primary method relied on to evaluate the benefits of dignity therapy and recommend it for clinical practice is self-report questionnaires, where high numbers of participants reported that dignity therapy was helpful to them, improved their sense of dignity, purpose and meaning, and where almost all participants reported dignity therapy to be a satisfactory experience (Chochinov, Kristjanson et al. 2011). Similar results on the feedback questionnaire were reported in the previous pilot study (Chochinov, Hack et al. 2005), and several studies have gone on to use the same or a very similar feedback questionnaire in their research, reporting similar results (Passik, Kirsh et al. 2004, Hall, Goddard et al. 2011, Chochinov, Cann et al. 2012, Hall, Goddard et al. 2012, Houmann, Chochinov et al. 2014). In addition, some dignity therapy studies examining potential benefits to family members have used a similar questionnaire with consistent results (McClement, Chochinov et al. 2007, Chochinov, Cann et al. 2012, Johns 2013). Findings based on these self-report measures are arguably the most important and have formed the empirical base for the effectiveness of dignity therapy. Some would argue that findings based on self-report are inherently unreliable (Nisbett

and Wilson 1977) but there is also a case to be made that self-report measures are as valid as other outcomes (Haeffel and Howard 2010).

Subsequent studies have attempted to strengthen these findings by using a variety of outcome measures to examine whether dignity therapy diminishes psychological distress. From this research, the most promising instrument appears to be the Herth Hope Index (HHI) (Herth 1990, Buckley and Herth 2004) assessing hopefulness as studies in the UK and Iran have both reported increased hope using this measure after dignity therapy (Hall, Goddard et al. 2011, Vaghee, Javadi et al. 2012). Another promising instrument is the Hospital Anxiety and Depression Scale (HADS) (Zigmond and Snaith 1983) as a result of a Portuguese study showing short-term improvements in anxiety and depression using this measure in cancer patients who had high levels of distress at baseline (Juliao, Oliveira et al. 2012). No other outcome measure used to date has been able to provide evidence of improved QOL or decreased psychological distress as a result of dignity therapy.

In their IRCT, Chochinov and colleagues suggested that low baseline levels of distress caused floor effects which prevented significant changes in the outcome measures (Chochinov, Kristjanson et al. 2011). The Portuguese study, which screened for high distress in participants and then showed reductions in anxiety and depression, provides support for this assertion (Juliao, Oliveira et al. 2012). However, a US study found people who were highly distressed had high dropout rates and were thus unable to benefit from dignity therapy (Johns 2013), and a UK case study examining the experiences of three distressed cancer patients concluded the complexities of delivering dignity therapy to distressed people may pose a substantial barrier (Hall, Goddard et al. 2013c). Thus, while dignity therapy may have the ability to decrease distress in distressed patients, it may not be feasible to perform dignity therapy with people experiencing substantial distress. Further research in this area is warranted.

Dignity therapy is based on an Chochinov's theoretical model of dignity in the terminally ill (Chochinov, Hack et al. 2002a), and it is designed to respond to the dignity concerns of role preservation, maintaining hope, maintenance of pride, continuity of self, generativity, care tenor, and aftermath concerns (Chochinov, Hack et al. 2005). Two

studies in the UK evaluated whether dignity therapy responded to the seven dignity related concerns it was designed to address (Hall, Goddard et al. 2013a, Hall, Goddard et al. 2013b). In the first study, through qualitative interviews with cancer patients in both the control and intervention groups, the researchers found that participants from both groups felt an increase in hopefulness, care tenor, maintenance of pride, and continuity of self as a result of participating in the research. Neither group reported improvements in aftermath concerns or role preservation, while the dignity therapy group alone experienced generativity benefits (Hall, Goddard et al. 2013b). The findings in a Danish study that people with children experienced more benefits from dignity therapy supports the idea that dignity therapy alleviates generativity concerns (Houmann, Chochinov et al. 2014).

Similar interviews assessing dignity related concerns were conducted with people in aged care in both the control and intervention groups. In this study, both groups reported that they received benefits from participating in research, but the study found no benefits relating to the seven areas in the theoretical model of dignity that dignity therapy is meant to address (Hall, Goddard et al. 2013a). These findings suggest that dignity therapy is most effective at addressing generativity concerns in people with terminal illness, and that person-centered contact, be it through an intervention or contact with researchers, provides numerous dignity-related benefits in and of itself (Chochinov 2007).

Initial steps have been taken to determine if dignity therapy can be used outside of palliative care settings. Three studies examined the application of dignity therapy with elderly people residing in aged care settings, which is a deviation from the palliative care population but also a group that faces the end of life and suffers from dignity-related distress (Chochinov, Cann et al. 2012, Hall, Goddard et al. 2012, Hall, Goddard et al. 2013a). Two of the studies reported more benefits (via the feedback questionnaire) in the intervention group than in the control group (Chochinov, Cann et al. 2012, Hall, Goddard et al. 2012), although the remaining study concluded that the excessive time needed to deliver the therapy, the negative experiences by family members, and the cognitive issues of residents indicate that dignity therapy may not be appropriate in this clinical setting (Hall, Goddard et al. 2013a).

Several studies have looked at other deviations from the dignity therapy intervention protocol (Chochinov 2012). The feasibility of conducting dignity therapy in aged care settings with people who suffer from dementia through a family member proxy was assessed. In this study, the researchers suggest dignity therapy conducted in this manner is an acceptable deviation offering benefits to all involved parties including the resident, family and staff (Chochinov, Cann et al. 2012). Novel applications of dignity therapy in people with chronic illness (Vaghee, Javadi et al. 2012), people with mental illness (Avery and Baez 2012) (Avery and Savitz 2011), and people in their homes using videoconferencing (Passik, Kirsh et al. 2004) appear to offer benefits but require further study. Dignity therapy may need to be modified to be performed with non-Western cultural groups. A Danish study found the question protocol required revision (Houmann, Rydahl-Hansen et al. 2010), and a Japanese study found dignity therapy is culturally incompatible (Akechi, Akazawa et al. 2012).

Research evaluating the impact of dignity therapy on family members substantiates initial findings that dignity therapy conducted with patients can be beneficial to family (McClement, Chochinov et al. 2007). However, in one study with family members of advanced cancer patients, two-thirds of family members also described negative experiences from dignity therapy (Hall, Goddard et al. 2013b), and family members of aged care residents also voiced worries and concerns about the therapy (Goddard, Speck et al. 2013) . These findings are very different from the findings of the IRCT, where Chochinov and colleagues (2011) reported "a few occasions family members were dissatisfied with the generativity document" (p. 761). Negative experiences were also reported in some aged care participants who felt the dignity therapy document created problems (Hall, Goddard et al. 2013a), which is different to Chochinov et al.'s pilot study and IRCT where no instances of dissatisfaction or distress were reported by dignity therapy participants (Chochinov, Hack et al. 2005, Chochinov, Kristjanson et al. 2011). Further research is warranted, including longitudinal studies, to investigate potential negative experiences and distress caused to both participants and family members as a result of the dignity therapy intervention and generativity document.

Four studies have examined the themes contained in dignity therapy transcripts (Hack, McClement et al. 2010, Montross, Winters et al. 2011, Tait, Schryer et al. 2011, Schryer, McDougall et al. 2012), although the approach in each of these studies was different making comparison difficult. One study assessed common values (Hack, McClement et al. 2010), two addressed common themes (Montross, Winters et al. 2011, Tait, Schryer et al. 2011), and another evaluated narrative types found in the documents (Schryer, McDougall et al. 2012). The findings of these studies suggests the material discussed in the documents is consistent with the questions asked in the dignity therapy question framework, including autobiographical information, important roles, family and relationships and accomplishments. Legacy messages, such as guidance and advice to family members, were also important themes. In two studies, a theme emerged that was not apparent in the question protocol, which was overcoming challenges and adversity (Montross, Winters et al. 2011, Tait, Schryer et al. 2011).

Finally, several studies addressed the practical realities of delivering dignity therapy. Overall, dignity therapy clinicians reported that delivering the therapy was a meaningful and worthwhile experience that increased their connection to patients (Montross, Meier et al. 2013, Tait and Hodges 2013, Davis Berman 2014). Health care providers of involved participants also reported positive impacts from the intervention, including increased knowledge about a patient as a result of reading their document (Chochinov, Cann et al. 2012, Montross, Meier et al. 2013). Three studies looked at pragmatic aspects of delivering the therapy, including time taken, costs expended and length of documents, but the findings are limited by incomplete information reported in these studies (Montross, Winters et al. 2011, Hall, Goddard et al. 2012, Johns 2013). Two studies noted that the time required to edit documents may be excessive and problematic for clinical implementation (Passik, Kirsh et al. 2004, Hall, Goddard et al. 2012), and one noted that travel time, cancelled appointments, and delays were common issues that compounded the substantial time commitment required by clinicians to complete the intervention (Hall, Goddard et al. 2012). Future studies that report comprehensively on the time and resources required to deliver dignity therapy are needed, especially in light of findings that person-centered contact alone appears to offer

many of the same benefits as dignity therapy (Hall, Goddard et al. 2013a, Hall, Goddard et al. 2013b).

## 3.5 Summary

MND presents numerous psychosocial and existential challenges for people diagnosed with the disease and their family carers throughout the disease trajectory. Research into significant psychosocial stressors on people with MND has shown that hopelessness, spirituality, and dignity are important areas of potential distress requiring attention and intervention. In MND family carers, anxiety and depression, caregiver burden, and hopelessness are important factors. In addition, the level of distress in either the person with MND or their family carer impacts on the level of distress of the other party. This shows that alleviating distress in one may have a beneficial impact for both. The impact of these challenges and stressors are more significant than the physical challenges encountered in MND on QOL. Because MND has no cure, maintaining QOL for people with MND and their caregivers is a primary treatment goal. However, despite the importance of psychosocial and existential distress on QOL, the vast majority of MND research is dedicated to the biological arena including the treatment of physical symptoms. Despite numerous calls in the literature, only one psychological, psychosocial, existential or spiritual intervention can be found in the literature to provide support or treat distress in people with MND or their family carers.

Dignity therapy was developed to alleviate psychosocial and existential distress at the end of life. It was designed to decrease suffering, enhance QOL, create spiritual peace, strengthen relationships, and increase a sense of meaning, purpose and dignity at the end of life through the creation of a legacy document. Approximately two dozen studies on various aspects of dignity therapy have built an empirical understanding of its benefits, but the effectiveness of dignity therapy has been difficult to fully document. Nonetheless, some studies have shown increases in hope and decreases in anxiety and depression as a result of dignity therapy, along with reports of increased meaning and decreased burden. Dignity therapy has proven feasible, but it may take longer to complete for people in aged care than for people with advanced cancer.

One of the most positive indicators of benefits derived from dignity therapy is that it has proven highly acceptable in all research settings, and participants and family carers report numerous benefits in self-report feedback questionnaires. Clinicians and health care professionals also report benefits from the therapy. On the other hand, although initial studies did not document negative experiences from the therapy, recent research highlights some problems with certain groups, including family carers hurt by some aspect of the document and aged care residents who do not feel the document is complete or worry about how it will be perceived. There has also been some challenge to the empirical basis for dignity therapy in findings that do not support the seven dignity related concerns among people receiving dignity therapy that dignity therapy is meant to address.

Nonetheless, dignity therapy is a promising psychosocial intervention to alleviate distress and enhance the end of life of people with MND and their family carers, but its feasibility, acceptability and potential effectiveness must be assessed. The experience of MND differs substantially from the experience of cancer, and cancer patients and family carers are the participants in most dignity therapy research, including the IRCT. Levels of distress differ in these two groups, as well as physical symptoms which may impact on the therapy. By exploring the use of dignity therapy with people who have MND, the three challenges confronting palliative care are being addressed, including researching a psychosocial intervention, performing research which impacts on the care and supports provided to family carers, and researching a palliative care intervention that impacts on a disease group other than cancer. This research supports the Dame Cicely Saunders grounding philosophy of palliative care to address the total pain encountered by people with MND and their family carers during their illness, and to help them live fully until death.

## CHAPTER FOUR
## 4. Methodology

Chapter Four consists of a published study protocol paper outlining the methodology of the study. The paper is entitled, "Is dignity therapy feasible to enhance the end of life experience of people with motor neurone disease and their family carers." It was published in the open access, peer-reviewed journal *BMC Palliative Care*. The design is a cross-sectional one group pre/post-test design. Outcomes for participants included hopefulness, spirituality/meaning and dignity. Outcomes for family carers included caregiver burden, hopefulness and anxiety/depression. Acceptability with both groups was to be determined through a feedback questionnaire, which was the same measure used in the IRCT.

> She said she is preparing me, because the results point to motor neurone disease. I was shocked but too numb to react. I don't want to think about it. …The occupational therapist came to assess our problems. She suggests we get a stair lift or a proper lift. What a bore. I felt depressed about what the future holds for me, and so sorry for poor Joss having this burden of my weakness, and all the expense of adapting the house for my illness. ...The therapist came. She was so depressing – she wanted wheelchairs and handrails and lifts. I found her forcefulness oppressive, and I want to escape. Joss and I can muddle on together – it's much better. …We had brill for supper. At the end of the meal I suddenly felt very tearful and could not stop weeping. My speech is getting slower and my right hand weaker. I am so frustrated and desperate. (Ackland and Ackland 2009 pp. 211-214)

Selected lines from a few pages of Rosemary Ackland's diary about being diagnosed with MND and the beginning of her MND journey. Rosemary's husband is British actor Joss Ackland, who published Rosemary's diary after her death from MND in 2002.

BMC Palliative Care

STUDY PROTOCOL Open Access

# Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers?

Brenda Bentley[1*], Samar M Aoun[1], Moira O'Connor[1], Lauren J Breen[2] and Harvey Max Chochinov[3]

## Abstract

**Background:** Development of interventions that address psychosocial and existential distress in people with motor neurone disease (MND) or that alleviate caregiver burden in MND family carers have often been suggested in the research literature. Dignity therapy, which was developed to reduce psychosocial and existential distress at the end of life, has been shown to benefit people dying of cancer and their families. These results may not be transferable to people with MND. The objectives of this study are to assess the feasibility, acceptability and potential effectiveness of dignity therapy to enhance the end of life experience for people with motor neurone disease and their family carers.

**Methods/design:** This is a cross-sectional study utilizing a single treatment group and a pre/post test design. The study population will comprise fifty people diagnosed with MND and their nominated family carers. Primarily quantitative outcomes will be gathered through measures assessed at baseline and at approximately one week after the intervention. Outcomes for participants include hopefulness, spirituality and dignity. Outcomes for family carers include perceived caregiver burden, hopefulness and anxiety/depression. Feedback and satisfaction with the intervention will be gathered through a questionnaire.

**Discussion:** This detailed research will explore if dignity therapy has the potential to enhance the end of life experience for people with MND and their family carers, and fill a gap for professionals who are called on to address the spiritual, existential and psychosocial needs of their MND patients and families.

**Trial registration:** ACTRN Trial Number: ACTRN12611000410954

**Keywords:** Motor neurone disease, Amyotrophic lateral sclerosis, Palliative care, Existential distress, Family carers, Dignity therapy

## Background

As a result of elevated interest in hastened death at the end of life by people with motor neurone disease (MND), also known as amyotrophic lateral sclerosis (ALS), numerous studies have examined factors that affect quality of life, psychological health, and end-of-life distress in this population. Findings indicate that quality of life in MND-diagnosed individuals is independent of physical decline [1,2]; that interest in hastened death is correlated with hopelessness [3,4]; and that MND patients with higher levels of spirituality and sense of meaning experience less end of life distress [5,6]. This research has resulted in a call to develop psychosocial interventions for use with the MND population that will bolster hopefulness, spirituality, and meaning [7,8]; however, very little work has been done to develop and implement such interventions.

MND is a family disease, and family carers carry an exceptional burden by providing a high level of care, often for the duration of the illness. Family carers of people with MND are more depressed than people with MND overall [9]. As time goes on and dependency increases, family carers exhibit increasing levels of distress symptoms [10,11]. Studies on the quality of life in

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MND family carers suggests that perceived caregiver burden can be alleviated by finding positive meaning [12,13] and by supporting a sense of hope [14].

Dignity therapy, a brief psychotherapeutic intervention based on empirical research into the concept of dignity at the end of life [15], has proven successful at increasing hope, sense of meaning and will to live in a palliative care population, where most patients had cancer diagnoses. Dignity therapy offers people with terminal illness the opportunity to create a generativity document. In a recorded interview guided by a counsellor or health care professional, the participant is invited to recount aspects of their life they want remembered, find meaning and purpose to their life, and express final words or advice. The interview is transcribed and edited, and a final dignity therapy transcript is returned to the participant to share with others as they wish.

A pilot study of dignity therapy produced positive results for participants and family members. A heightened sense of dignity was reported in 76% of participants, an increased sense of purpose was reported by 68%, an increased sense of meaning was reported by 67%, and 47% reported an increased will to live [16]. A recent randomised controlled trial reported similar outcomes [17]. In the pilot study, family members were also positive about the intervention, with 95% reporting they would recommend dignity therapy, 78% reporting that it helped them during their time of grief, and 77% believing the document would be a continuing source of comfort [18].

Nonetheless, because these studies both utilized a primarily cancer population, the results are not transferable to people with MND. Ability to communicate, cognitive acuity, stage of illness, baseline levels of distress and demographic features are some of the factors that may vary in this population and make implementation of dignity therapy difficult. The delivery of the intervention may require modification, i.e. to be performed at an earlier stage in the disease process or by utilizing assisted communication methods. Therefore, feasibility testing of dignity therapy with the MND population is warranted [19,20].

Moreover, while the previous dignity therapy study focused on the intervention's positive influence on the bereavement experience of family members, it did not look at how the intervention may affect the carer during the caring experience.

### Aims and objectives

The aims of this study are to assess the feasibility, acceptability and potential effectiveness of dignity therapy to enhance the end of life experience for people with MND and their family carers. The specific objectives are to:

a) Determine whether dignity therapy is likely to increase hope, meaning and dignity in people with MND.
b) Determine whether dignity therapy is likely to increase hope, and decrease anxiety, depression, and perceived burden in family carers of people with MND.
c) Determine whether dignity therapy is acceptable to people with MND and their family carers.
d) Determine whether it is feasible to provide dignity therapy to people with MND.
e) Pilot methods for a future randomized controlled trial.

## Methods

### Study design

This is a cross-sectional study utilizing a single treatment group and a repeated measures pre/post test design. A control group is not being utilized due to 1) the small MND population available in Western Australia 2) access issues to people with MND, and 3) the fact that dignity therapy (nor any other psychosocial intervention) has yet to be tried with this palliative care population and the feasibility of dignity therapy needs to be tested before proceeding to an RCT. The study design has been modelled to reduce bias and increase validity where possible. For example, the short duration of the intervention coupled with utilizing a single post-testing point one week after the intervention will minimize confounding variables.

### Ethical approval

This study has been approved by the Curtin University Human Research Ethics Committee (19/2011).

### Participants

The sample will comprise 50 adults diagnosed with MND who are registered with the MND Association of Western Australia. A second group will consist of up to 50 family carers of the participants.

#### *People with MND*

**Inclusion criteria** Persons with a diagnosis of MND aged 18 and over who are able to communicate in English are included in the study. As this is a feasibility study, participants who are unable to communicate verbally may participate if they are able to utilize an assisted communication method. Communication issues will be explored and reported in the findings. There is no selection criteria based on stage of the disease as MND is a fatal disease with no hope of remission making an end-of-life intervention appropriate at any time after diagnosis. Participants will not be screened for existential or

psychosocial distress; however, these will be assessed at baseline.

**Exclusion criteria** Participants who are unable to provide informed consent, either due to cognitive issues or illness severity will be excluded. Cognitive impairment will be screened using the ALS-Cognitive Behavioral Screen [36]. If the participant receives a score of less than ten, the participant and family carer will be excluded from the study.

#### *Family carers*

A family carer of each participant will be invited to take part in the study, comprising a second research population of up to 50 family carers. A family carer is defined as the person indicated by the participant as the primary family carer. The family carer must be at least 18 years of age, able to provide informed consent and able to communicate in English. If the family carer does not wish to participate, then the person with MND remains eligible to take part in the study.

### The intervention and study procedures

The intervention will be administered by the researcher, a counselling psychologist undertaking a PhD by research at Curtin University in Western Australia. The researcher has been trained in dignity therapy at an intensive workshop by Professor Harvey Chochinov, who developed the intervention and who performed the empirical studies upon which the intervention is based.

Recruitment will be undertaken through the MND Association of Western Australia (MNDAWA). MNDAWA will liaise directly with participants to protect the identity of MND-diagnosed individuals. Letters will be sent out to all members of MNDAWA and potential participants are identified when they telephone the researcher or return a copy of the letter expressing their interest. The researcher will first speak with the person with MND or family carer to elaborate further on the nature of the study and answer any questions. If the participant wishes to proceed, a meeting will be scheduled. If time permits, information sheets and consent forms will be mailed in advance of the meeting to provide time to consider the study and discuss their participation with family and health care providers. All meetings will occur at a time of the participant's choosing and in their care environment. Since the participant's condition can fluctuate, the timing of the contacts can be flexible and meetings rescheduled. The researcher will make detailed notes of these experiences in order to include information about timing issues in the findings.

At the initial meeting, the researcher will review the participant and family carer information and consent forms with the participant/family carer dyad, check for understanding and answer any outstanding questions. Written consent will be obtained and MND participants will be tested for cognitive impairment that is significant enough to exclude them from the study. If the participant and family carer remain in the study, socio-demographic and health questionnaires and baseline measures will be collected from each. An appointment for the first dignity therapy session will be made, ideally within two to three days. The researcher will provide the participant a copy of the dignity therapy question framework so that they may begin reflecting on their responses.

Dignity therapy question protocol

> Tell me a little about your life history; particularly the parts that you either remember most or think are the most important? When did you feel most alive?
>
> Are there specific things that you would want your family to know about you, and are there particular things you would want them to remember?
>
> What are the most important roles you have played in life (family roles, vocational roles, community service roles, etc.)? Why were they so important to you, and what do you think you accomplished in those roles?
>
> What are your most important accomplishments, and what do you feel most proud of?
>
> Are there particular things you feel still need to be said to your loved ones or things that you would want to take time to say once again?
>
> What are your hopes and dreams for your loved ones?
>
> What have you learned about life that you would want to pass along to others? What advice or words of guidance would you wish to pass along to your son, daughter, husband, wife, parents, other(s)?
>
> Are there words or perhaps even instructions that you would like to offer your family to help prepare them for the future?
>
> In creating this permanent record are there other thing you would like included?
> [16] at p. 5522.

The next meeting is the primary dignity therapy session, where participants are invited to address and record themes, thoughts and feelings about themselves and their lives that they would like remembered. The question framework is flexible, and provides a guide for the researcher to shape the interview by following the participant's cues. If the participant desires, a family carer

may be present during the interview to provide emotional support and help facilitate the interview. If participants require a second or third interview session due to communication issues, fatigue, or the breadth of information they would like to share, these will be scheduled as soon as possible.

After recording has finished, a verbatim transcript will be prepared (usually within 24–48 hours). The researcher will shape the interview into a narrative using the learned dignity therapy editing method, which omits non-starters and irrelevant sections (such as interruptions) and tags content that needs to be clarified or perhaps omitted due to the harm it could inflict on recipients. Another appointment will be made with the participant as soon as practicable to read through and edit the transcript. In this session, the participant will be invited to make corrections, clarifications or additions as desired. In the final dignity therapy session, the final bound transcript is given to the participant. The participants may have as many copies of the document as they wish and share them with whoever they choose.

Post-testing will occur with both the participant and family carer one week after the final dignity therapy document has been returned. To reduce response bias, post-testing questionnaires will be sent out and returned via mail. A research officer (not related to the project) from the Western Australian Centre for Cancer and Palliative care will perform the post-testing if assistance is needed to complete the questionnaires. A single point of post-testing soon after completion of the sessions is planned in order to reduce moderating variables. Further, the possible immediate impact of the intervention is being examined for the purposes of this feasibility study and not the longevity of the outcomes, which can be assessed in future studies. A study design flow chart is shown in Figure 1.

## Measures and outcomes

The measures chosen have been validated and are quick and easy to administer and use, in order to decrease the burden on the study population. Brief versions of the outcome measures have been selected where available.

### *Primary outcome measure for people with MND*

The primary outcome measure is the participant's sense of hopefulness. This will be assessed utilizing the Herth Hope Index [21,22], a validated instrument developed for use with the terminally ill. Distressed MND patients score higher on hopelessness than distressed cancer patients at the end of life [23], and interest in hastened death or suicidal ideation in the MND population has been shown to correlate with hopelessness [3,24], which indicates that the level of hopefulness a key factor of psychological distress in this disease population.

### *Secondary outcome measures for people with MND*

Secondary outcome measures used to assess potential effectiveness include, 1) dignity, which will be measured with the Patient Dignity Inventory (PDI) [25], a validated measure which evolved directly from the empirical studies into dignity concerns in the terminally ill, and; 2) spiritual well-being, which will be measured with the Functional Assessment of Chronic Illness Therapy-Spiritual Well-Being Scale (FACIT-sp-12) [26], a validated measure showing strong internal reliability. To assess the acceptability of dignity therapy, a modified version of the dignity therapy Patient Feedback Questionnaire [17] is being utilized to collect information on the views, experiences and opinions of the participants about the intervention. To assess the feasibility of the intervention, data will be collected about the time taken to organize and conduct the dignity therapy sessions, any special accommodations made in the delivery of the intervention, deviations from the dignity therapy protocol, and reasons for non-completion. The researcher will also record, through the use of a journal, observations and experiences of delivering the intervention, including positive and negative participant responses.

### *Primary outcome measure for family carers*

The primary outcome is the family carer's sense of perceived burden, which will be measured through the Zarit Burden Inventory [27]. Research of caregiver burden in MND family carers has documented the considerable burden attached to caring for persons with MND [28,29]. Caregiver burden increases as patient function declines [30-32]. Studies indicate that MND caregiver burden can be alleviated by finding positive meaning [12,13] and by supporting a sense of hope [14]. It can be inferred that dignity therapy may impact hope and meaning, thereby alleviating caregiver burden. In the alternative, whether this intervention increases burden is a factor that also must be considered when evaluating its overall impact.

### *Secondary outcome measures for family carers*

Secondary outcome measures used to assess the potential impact of dignity therapy on family carers includes 1) hopefulness, utilizing the Herth Hope Index [21]. The HHI has been used successfully in studies with family caregivers of terminally ill patients [33], and 2) anxiety and depression, which will be measured with the Hospital Anxiety and Depression Scale [34], an instrument often used with family caregivers showing strong reliability and validity. To assess the acceptability of dignity therapy to family carers, a modified version of the dignity therapy Family Feedback Questionnaire [17] is being utilized to collect information on the views, experiences and opinions of the family carers about the intervention.

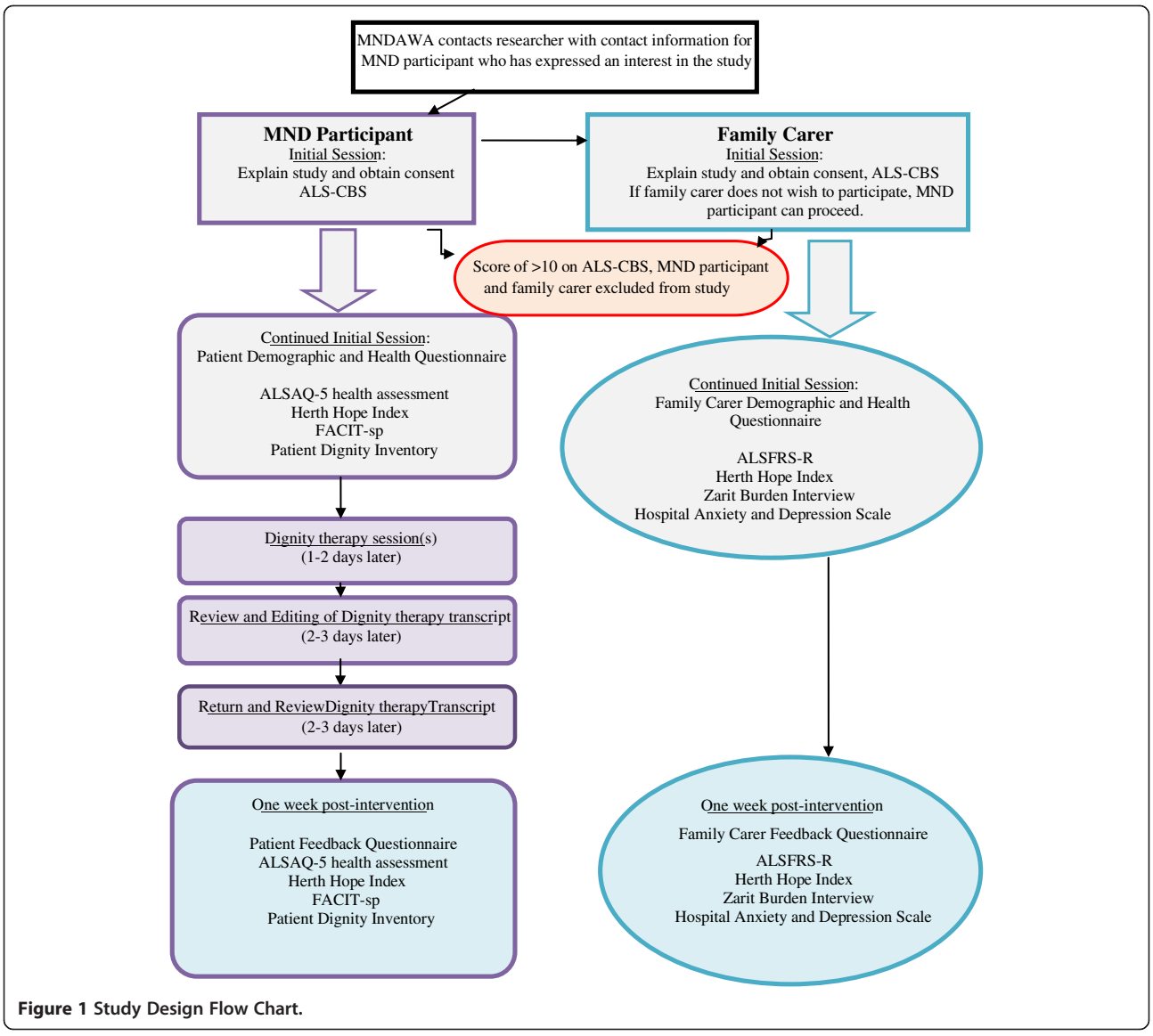


**Figure 1** Study Design Flow Chart.

### *Demographic and health status*

Demographic and health status information collected from persons with MND will include disease specific health-related quality of life utilizing the Amyotrophic Lateral Sclerosis Assessment Questionnaire-5 [35], cognitive behavioural functioning from the carer's perspective utilizing the ALS Cognitive Behavioral Screen [36], age, gender, and health history.

Demographic and health status information collected from family carers will include the level of disability of the care recipient utilizing the Amyotrophic Lateral Sclerosis Functional Rating Scale-R [37,38], cognitive behavioural functioning from the carer's perspective utilizing the ALS Cognitive Behavioral Screen [36], age, gender, relationship to the person with MND, caring hours per day, employment status, and health history.

## Analysis

For summarizing purposes, descriptive statistics will be obtained for demographic variables. To assess the possible impact of the intervention on the psychosocial and existential concerns of the participant and family carer, pre and post intervention comparisons for each outcome variable will be carried out using Wilcoxon's signed rank-sum tests, given that the main outcome variable measured will not be normally distributed [16]. Spearman's rank correlation coefficient will be calculated to assess the possible correlation between variables of interest. It is anticipated that there will be a post-intervention improvement on all psychosocial measures for both the participant and family carer after the dignity therapy intervention. Open-ended responses in feedback questionnaires will be coded and analysed using

descriptive statistics. The IBM SPSS version 20 statistical software package will be used for all analyses. A p value less than 0.05 is considered to be statistically significant.

## Discussion

Several studies, as well as MND practice guidelines, suggest the need to develop and utilize interventions that will support hopefulness, a sense of meaning, and dignity in order to alleviate psychosocial and existential distress in persons with MND [7,8]. Despite this, very little has been done to develop or implement such interventions. In fact, an extensive literature search completed for this study revealed there were no psychosocial interventions specifically designed or tailored to alleviate existential distress and improve the quality of life of persons with MND. This research will begin to fill this gap, providing a possible solution to a concern about this specific population, as well as continue to advance the overall focus of alleviating psychosocial distress at the end of life, an area of palliative care which has been widely acknowledged as being in need of improvement.

This study will determine if dignity therapy is likely to be effective in enhancing the end of life experience for both people with MND and their family carers, if it is acceptable to people with MND and their family carers, and if it is feasible to offer the intervention to this population. It will explore what allowances or modifications might need to be made to deliver the intervention to people who are sometimes unable to communicate verbally. It will provide a preliminary examination of how cognitive or neurobehavioral issues encountered in persons with MND might affect the intervention, including the completion and sharing of a generativity document that reflects a true sense of the person with MND. Finally, if the intervention is unsuccessful, this study will determine what factors contributed to a negative outcome, as well as any unexpected consequences to persons with MND or their families.

This research has implications for psychological and health care professionals who work with people with motor neurone disease and other neurological disorders, as well as those who work in palliative care settings. Both are often called on to address the spiritual, existential and psychosocial needs in their patients. If dignity therapy proves to be effective, it could be a relatively brief and easy to administer intervention that could be made available to people with MND. This study has the potential to provide a precise intervention to ameliorate psychosocial and existential distress, as well as improve the quality of care provided to people with MND and their family carers.

#### Competing interests

The authors declare they have no competing interests.

#### Authors' contributions

SA and HC conceived the study. BB, SA, MO and HC designed the study utilizing methods developed by HC in previous dignity therapy pilot and RCT studies. BB drafted the article and will carry out the research. SA, MO, LB and HC will supervise the research. HC developed dignity therapy. All authors made substantial contributions to the critical revision of the article and approved the final content.

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## CHAPTER FIVE
## 5. Study findings relating to people with MND

Chapter Five consists of a published manuscript of the study findings relevant to people with MND. The paper is entitled, "Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease." This paper was published in the open access, peer-reviewed interdisciplinary journal *PLoS One*. The results of the study on 29 people with MND showed no significant changes on the outcome measures on the group level, but there were increases in hope on the individual level. Dignity therapy was acceptable and the results of the feedback questionnaire were similar to the results found in the IRCT. Dignity therapy is feasible if the therapist can overtime time and communication difficulties.

> I'm a different self now. I'm different in my attitudes. I'm different appreciating my body, which I didn't do fully before. I'm different in terms of trying to grapple with the big questions, the ultimate questions, the ones that won't go away.
>
> And which are the important questions?
>
> As I see it, they have to do with love, responsibility, spirituality, awareness. And if I were healthy today, those would still be my issues. They should have been all along. (Albom 1997 p. 175)

A conversation between Morrie Schwartz, a retired professor and a man with MND, and Mitch Albom, his former student and journalist. Albom wrote the memoir "Tuesdays with Morrie" about 14 weekly meetings/interviews he had with Schwartz in the final months of his life. The book became a bestseller and was made into a movie starring Jack Lemmon as Morrie.

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# Feasibility, Acceptability, and Potential Effectiveness of Dignity Therapy for People with Motor Neurone Disease

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## Abstract

***Background:*** Motor neurone disease (MND) practice guidelines suggest developing interventions that will promote hope, meaning, and dignity to alleviate psychological distress, but very little research has been done. This study begins to address this need by exploring the use of dignity therapy with people with MND. Dignity therapy is a brief psychotherapy that promotes hope, meaning and dignity, and enhances the end of life for people with advanced cancer. The aims of this study are to assess the feasibility, acceptability, and potential effectiveness of dignity therapy for people with MND.

***Methods/design:*** This cross-sectional feasibility study used a one-group pre-test post-test design with 29 people diagnosed with MND. Study participants completed the following self-report questionnaires: Herth Hope Index, FACIT-sp, Patient Dignity Inventory, ALS Assessment Questionnaire, ALS Cognitive Behavioural Screen, and a demographic and health history questionnaire. Acceptability was measured with a 25-item feedback questionnaire. Feasibility was assessed by examining the length of time taken to complete dignity therapy and how symptoms common in MND affected the intervention. Generalised linear mixed models and reliable change scores were used to analyse the data.

***Results:*** There were no significant pre-test post-test changes for hopefulness, spirituality or dignity on the group level, but there were changes in hopefulness on the individual level. The results of the feedback questionnaire indicates dignity therapy is highly acceptable to people with MND, who report benefits similar to those in the international randomised controlled trial on dignity therapy, a population who primarily had end-stage cancer. Benefits include better family relationships, improved sense of self and greater acceptance. Dignity therapy with people with MND is feasible if the therapist can overcome time and communication difficulties.

***Conclusions:*** Dignity therapy for people with MND is feasible and acceptable. Further research is warranted to explore its ability to diminish distress.

***Trial Registration:*** www.anzctr.org.au ACTRN12611000410954

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## Introduction

Motor neurone disease (MND), also known as amyotrophic lateral sclerosis, is an uncommon neurodegenerative disease that is progressive and always fatal. There is no cure and few options exist for treatment. While a few die within six months, others live ten years or more. On average, people live two to three years after diagnosis before eventually succumbing to paralysis and death, most often from respiratory failure [1,2].

Despite the considerable physical and emotional suffering involved, there is little focus on addressing the psychological needs of people with MND. Quality of life is generally high [3–5], but people with MND often experience significant psychological distress including anxiety and hopelessness [6]. Psychological distress in MND is associated with decreased quality of life and decreased survival rates [7,8]. Hopelessness is correlated with interest in hastened death [3,9,10]. Among those with terminal diagnoses, people with MND report the highest levels of interest in hastened death [11] and they also have the greatest risk of suicide [12]. These effects are mitigated in people who report higher levels of spirituality and sense of meaning [13,14]. Such findings have led to calls for psychological interventions to bolster hopefulness, spirituality, and meaning in people with MND [1,15]; however, intervention studies are lacking.

One promising intervention is dignity therapy; a brief psychotherapeutic intervention based on an empirical understanding of dignity at the end of life [16]. Dignity therapy offers people facing death the opportunity to create a document about their life [17]. In a recorded life reflection interview, a person with terminal disease is afforded the opportunity to discuss significant memories, meaningful events and important accomplishments, as well as leave messages for loved ones. In previous studies, dignity therapy

has been shown to alleviate existential distress in a palliative care population where most people had malignant conditions [18,19]. With a key aim to bolster hope and meaning, dignity therapy has the potential to alleviate psychological distress in people with MND [15]. However, because most of the people in previous dignity therapy research had terminal cancer, the findings of its effectiveness are not transferable to people with MND. Diagnosis, ability to communicate, cognitive acuity, stage of illness, baseline levels of distress and demographic features are factors that differentiate people with MND from people with end-stage cancer. Finally, delivery of dignity therapy to people with MND may require modification, for example, to be performed at an earlier time or via assisted communication methods.

### Aims and objectives

The aim of this study was to assess the feasibility, acceptability, and potential effectiveness of dignity therapy to enhance the end of life experience for people with MND. The specific objectives were to determine whether:

a) dignity therapy increases hope, meaning, and dignity in people with MND;
b) dignity therapy is acceptable to people with MND; and
c) it is feasible to provide dignity therapy to people with MND.

## Methods

### Study design

This cross-sectional study utilized a one-group pre-test-post-test design. A control group was not utilized due to 1) the small MND population, 2) access issues to people with MND, 3) ethical concerns over making a potentially useful intervention unavailable to a control group, and 4) the need to test the feasibility of dignity therapy with people with MND [20,21]. Further details can be found in our protocol [22].

### Ethical approval

This study was approved by the Curtin University Human Research Ethics Committee (19/2011).

### Setting

Participants were primarily enrolled as a result of outreach from the Motor Neurone Disease Association of Western Australia (MNDAWA). MNDAWA sent recruitment letters to people who had been diagnosed with MND and referred to their services by a general practitioner or neurologist. In the last six months of the study, we used social networking, a press release, and information on the university web site to assist in reaching the recruitment goal. One participant in Queensland participated via video-conferencing. Twenty-two participants reported living in an urban/metropolitan area and seven in rural areas. Twenty-seven participants were living at home at the time of the intervention, one in an aged-care facility and one a hospital.

### Participants

Individuals diagnosed with MND, over 18 years old, who could communicate in English and provide informed consent (based on the ALS-Cognitive Behavioural Screen (ALS-CBS) [23] where a cut-off score of 10 was used or the Blessed Orientation Memory Concentration (BOMC) test [24] where a cut-off score of 9 was used) were eligible for the study. Participants were provided with information sheets and written consent was obtained. Enrolment occurred between June 2011 and July 2013. People were excluded if they were too ill to complete the requirements of the protocol. There were no selection criteria based on distress levels, disease stage or proximity to death.

### The intervention

The intervention was administered by a researcher trained in dignity therapy by Harvey Max Chochinov who developed the therapy [18,19]. The therapy interviews were audio-recorded and transcribed verbatim by a transcriptionist. The researcher shaped the transcribed interviews using the prescribed editing process [17] and then returned to edit and complete the transcripts with the participants. The document was read aloud to each participant at the conclusion of the intervention. To mitigate response bias, post-testing took place via mail or through a visit from a second researcher. The researcher engaged in regular supervision sessions from Prof. Chochinov. To optimize adherence to the dignity therapy protocol, three recordings, transcripts, and completed documents (10%) were reviewed by three experienced researchers (two trained in dignity therapy) and deemed to be adherent.

### Measures and Outcomes

**Effectiveness.** Outcome data to measure potential effectiveness were collected from participants at baseline and one week after completion of dignity therapy. The primary outcome measure was the participant's sense of **hopefulness** assessed with the Herth Hope Index [25,26], a reliable ($\alpha = 0.97$) validated instrument developed for use with the terminally ill, with a score ranging from 12–48 and where higher scores indicate more hopefulness. Secondary outcomes were: 1) **Dignity,** measured by the Patient Dignity Inventory (PDI) [27]. The PDI has a scale of 25–125 (higher scores indicate greater distress). It is a reliable ($\alpha = 0.93$) validated measure which evolved directly from the empirical studies into dignity concerns in the terminally ill. [27] 2) **Spiritual well-being,** measured by the Spiritual Well-Being subscale of the Functional Assessment of Chronic Illness Therapy scale (FACIT-sp-12) [28]. The FACIT-sp-12 has a scale of 0–48 with higher scores indicating greater spiritual wellbeing, and it is a reliable ($\alpha = 0.87$) and valid measure [28].

**Acceptability.** The Participant Feedback Questionnaire used in the international randomised controlled trial of dignity therapy (IRCT) [19] was modified by adding three items on hopefulness and family support, and was used to collect the participants' experiences and opinions of the intervention. The questionnaire contained 25 questions answered with a 5-point Likert scale and space for brief explanation.

**Feasibility.** Data were collected about the time taken to conduct the therapy sessions, any special accommodations made in the delivery of the intervention, deviations from the dignity therapy protocol, reasons for non-completion, and reasons for attrition.

**Demographic and health status.** Disease specific health-related quality of life was measured with the Amyotrophic Lateral Sclerosis Assessment Questionnaire-5 (ALSAQ-5) where scores range from 0–20 (higher scores indicating more impairment) [29], and cognitive behavioural functioning was assessed with the ALS – CBS [23]. Level of impairment of the person with MND and change in physical function over time was collected from the family carer using the Amyotrophic Lateral Sclerosis Functional Rating Scale-R (ALS-FRS) where scores range from 0–48 (lower scores indicating more impairment) [30,31]. Demographic data on age, gender, education level, marital status, and health history were also collected.

### Analysis

Data were analysed with generalised linear mixed models (GLMM) as implemented through SPSS's (Version 20) GENLIN-MIXED procedure. Model parameters were estimated with robust standard errors in order to accommodate potential violations of the model assumptions. Participant was treated as a random effect and Time (pre-test, post-test) was treated as a fixed effect. Age, gender, time since diagnosis, marital status, level of education, and number of days from pre-test to post-test were also treated as fixed effects and analysed individually as potential moderators of the intervention effect. In order to optimise the likelihood of convergence, a separate GLMM analysis was run for each of the three outcome measures. The GLMM maximum likelihood procedure is a full information estimation procedure that uses *all* the data present at *each* assessment point. *All* of the pre-test data and *all* of the post-test data are incorporated into the analysis, which reduces sampling bias associated with participant attrition. GPower (Version 3.1) indicated that 29 participants would be sufficient to capture 'moderate to large' ($f = .28$) pre-post changes on the outcome variables. A reliable change (RC) score for each participant [32] was computed to investigate the presence of reliable pre-post change at the individual rather than group level. The RC score is the degree to which the person changes on the outcome variable divided by the standard error of difference between the pre- and post-test scores. When the absolute value of the RC score is greater than 1.96, (Wise [33] has argued that this value can be reduced in some situations), it is likely that the post-test score reflects a *real* or *reliable* change. Descriptive statistics were used to summarize demographic variables and feedback responses.

## Results

### Response rate

MNDAWA distributed recruitment letters to all 147 members diagnosed with MND on three occasions between May 2011 and May 2013. Thirty-five people responded (response rate 24%) and 29 of these people completed the study (completion rate 78%). Those who did not complete include three people who changed their mind before entering the study, two who changed their mind after entering the study, two who died before completion and one who was excluded due to cognitive impairment. While all 29 completed dignity therapy, one did not complete any post-test measures due to illness, one completed the feedback questionnaire but not the outcome measures, and three additional participants did not complete the PDI fully.

### Demographic information

Participants, 20 men and 9 women, ranged from 32 to 81 years of age with almost half between the ages of 60 and 69. Twenty-four were married or partnered. Thirteen reached secondary education; 16 achieved university or postgraduate education. (See Table 1 for more demographic information on the study population).

### Baseline levels of impairment and distress

The sample group was moderately impaired (ALS-FRS mean = 32.61, SD = 9.76). Scores on the ALSAQ-5 indicate the sample had moderate health-related quality of life (mean = 9.31, SD = 3.96). The group was hopeful, had low dignity-related distress, but appeared to be facing some struggles with their spiritual wellbeing (see Table 2 Pre-test scores). The mean total score for spiritual wellbeing was 30.7 (SD = 10.43) which was lower than in people with cancer (mean = 38.5, SD = 8.1) [28].

### Effectiveness

Descriptive statistics for the outcome variables are reported in Table 2. There were no significant pre-test post-test changes for hopefulness ($F$ [1,54] = 2.79, $p = .101$, $d = .46$), dignity ($F$ [1,54] = 0.45, $p = .504$, $d = .20$), or spirituality ($F$ [1,54] = 0.01, $p = .936$, $d = .05$). Potential moderators of the intervention effect (age, gender, time since diagnosis, marital status, level of education, and number of days from pre-test to post-test) were individually entered in the regression model in order to determine whether significant pre-test-post-test changes would be observed at certain values of the moderator. There was no significant Moderator x Time interactions for any outcomes (all $p$s $>.1$).

A reliable change (RC) score for each participant [32]. The results indicate that some individuals showed an improvement in hopefulness, while a quarter showed deterioration (see Table 3). Interestingly, all of the study participants who had an increase in hopefulness reported they were both religious and spiritual, while 43% of the group whose hopefulness declined reported they were neither religious nor spiritual. Additionally, 50% of the group with improved hopefulness had been diagnosed with MND for four years of more, while 85% of the group that declined had been diagnosed for two years or less.

### Acceptability

The participants found dignity therapy to be satisfactory (92.8%), helpful to them (89.2%), helpful to their family (85.2%), and would recommend dignity therapy to others with MND (84%). They reported the strongest positive improvements in the dignity-related areas of looking after unfinished business (67.9%), continuity of self (67.9%) acceptance (64.2%), and role preservation (60.8%). There were lesser improvements in feeling like a burden (28.6%), increased will to live (33.3%), lessened sadness or depression (35.7%), and sense of control (35.7%). Seventy percent reported they felt closer to the people who meant the most to them after dignity therapy, and 63% felt dignity therapy would result in better appreciation in them from their families.

The results of the feedback questionnaire are very similar to the results of the dignity therapy arm in the IRCT which showed that dignity therapy outperformed standard care in a palliative care population where 96% suffered from end-stage cancer [19] (see Table 4). In both studies, people undergoing dignity therapy reported the psychotherapy was helpful to them, improved their quality of life and increased meaning. These findings demonstrate that people with MND experience similar benefits from dignity therapy as reported in previous research with people with cancer [19] (see Table 4).

### Feasibility

Dignity therapy for the sample took from three to seven sessions, consistent with the standard protocol [17,34]. The majority of participants (69%) finished the therapy in four sessions (mean = 4.14). Four participants (13.8%) completed in the standard of two weeks [19]. The time to completion ranged from 7 to 152 days, with about half completing by 25 days (mean = 42, $SD = 36$). Reasons for extended completion times included (often in combination) the participants' speech impairment, travel, hospital or respite care admissions, family and employment obligations, and desire for more time to work on the document [35].

Participants' use of various assisted communication methods meant that dignity therapy was successfully completed with six people who, due to MND, had lost the ability to speak. An additional three people had moderate speech impairment and these participants had the three longest completion times (87, 134,

**Table 1.** Demographic characteristics of study group.

| | |
|---|---|
| **Gender** | |
| Male | 20 |
| Female | 9 |
| Age | |
| 30–39 | 1 |
| 40–49 | 1 |
| 50–59 | 4 |
| 60–69 | 15 |
| 70–79 | 6 |
| 80–89 | 2 |
| Marital Status | |
| Married | 24 |
| Widowed | 3 |
| Divorced/separated | 1 |
| Never married | 1 |
| Residence area | |
| Urban/metropolitan | 22 |
| Rural | 7 |
| Residence type | |
| Home | 27 |
| Hospital | 1 |
| Aged-care facility | 1 |
| Presently living with | |
| Spouse | 23 |
| Alone | 4 |
| Other | 2 |
| Highest level of education attained | |
| Secondary/high school | 13 |
| University/technical | 13 |
| Postgraduate | 3 |
| Current employment status | |
| None | 22 |
| Full-time | 2 |
| Part-time | 3 |
| On leave | 2 |
| Time since diagnosis | |
| Less than one year | 8 |
| One to two years | 9 |
| Two to three years | 4 |
| Three to four years | 0 |
| More than four years | 8 |
| Time since initial symptoms | |
| Less than one year | 2 |
| One to two years | 10 |
| Two to three years | 5 |
| Three to four years | 3 |
| More than four years | 9 |
| Prior history of depression (before MND diagnosis) | |
| Yes | 6 |
| No | 23 |
| Have you been prescribed medication to help you cope? | |

**Table 1.** Cont.

| | |
|---|---|
| **Gender** | |
| Anti-depressant | 7 |
| Anti-anxiety | 2 |
| Sleeping medication | 1 |
| Anti-anxiety & sleeping medication | 1 |
| No | 18 |
| Do you consider yourself to be a religious person? | |
| Yes | 8 |
| Somewhat | 10 |
| No | 11 |
| Do you consider yourself to be a spiritual person? | |
| Yes | 10 |
| Somewhat | 14 |
| No | 5 |
| Cognitive screening scores | |
| No impairment | 19 |
| Suspected mild to moderate impairment | 10 |

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and 152 days). One participant with moderate speech impairment completed the intervention using videoconferencing and email.

## Discussion

This is the first study to explore the feasibility of dignity therapy with people with MND and, to our knowledge, the first study of a targeted psychotherapeutic intervention for this population. We expected to detect measureable post-intervention increases in hope, dignity and spirituality at the group level but this did not occur. This may be due to a number of reasons, including the difficulties with demonstrating psychosocial change at the end of life with self-report measures [36], the result of evidence which suggests the benefits of psychosocial interventions at the end of life can most readily be shown in patients who have elevated levels of distress [37], and/or that the outcome measures chosen were not sensitive to the impacts that occurred. Very small pre-post effects were present for dignity and spirituality, and the effect for hopelessness was small to moderate. However, without a control group, we were unable to ascertain whether the intervention had a prevention effect against expected declines in hope, dignity, and spirituality over time as a person with MND deteriorates and approaches death. At the individual level, tentative findings are that dignity therapy may be effective at increasing hopefulness in people who are more spiritual and also in some with advanced disease, as reported in previous research [38].

Nonetheless, the positive results on the feedback survey indicate most people with MND believe dignity therapy to be beneficial. The intervention was found to be overwhelmingly positive. Feedback indicated dignity therapy helped enhance the end of life by supporting the unique identity of the person, helping with acceptance, allaying aftermath concerns, finding meaning and purpose, and improving family relationships which mirrors the previous findings of the pilot study and international randomised controlled trial of dignity therapy performed with people with end-stage cancer [18,19]. Moreoever, people with MND believe dignity therapy will be of help to their family members after death indicating a potential benefit to family members during bereavement as found in other studies [39,40]

### Feasibility

Dignity therapy with people with MND is feasible if the therapist can overcome time and communication difficulties, as it takes longer to administer with people who have MND than those with cancer. Therapist time was increased in order to travel to participants in their homes to deliver the intervention (previously completed in palliative care settings [19,34]) and was compounded for participants in rural areas. Ninety-three percent of participants were in the community rather than an in inpatient or care facility. As such, we were less in control of the schedule. Additionally, for people with speech impairment, dignity therapy was prohibitively difficult and time consuming to perform. Adapted methods appear

**Table 2.** Mean Pre-test Post-Test Scores on Measures for Hopefulness, Dignity, and Spirituality.

| Outcome | Pre-test | *N* | Post-test | *N* |
|---|---|---|---|---|
| Hopefulness (HHI) | 38.76 (5.10) | 29 | 36.61 (6.80) | 27 |
| Dignity (PDI) | 48.59 (15.45) | 29 | 47.59 (12.91) | 24 |
| Spirituality (FACIT-sp-12) | 30.72 (10.43) | 29 | 30.92 (9.88) | 27 |

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**Table 3.** Percentage (Number) of Participants Showing Reliable Improvement, Deterioration, and No Change for Hopefulness, Dignity, and Spirituality.

| Outcome | Improved | Deteriorated | No change | *N* |
|---|---|---|---|---|
| Hopefulness (HHI) | 14.8 (4) | 25.9 (7) | 59.3 (16) | 27 |
| Dignity (PDI) | 0 | 0 | 100 (24) | 24 |
| Spirituality (FACIT-sp-12) | 0 | 0 | 100 (27) | 27 |

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to present viable solutions to these issues, such as some of the therapy being conducted via email or utilizing videoconferencing.

Unique aspects common to MND, including speech impairment and mild to moderate cognitive impairment, did not detract from the benefits of the therapy. These results indicate that dignity therapy is feasible and acceptable, and it offers potential benefits for people with MND.

## Strengths and limitations

The strengths of this feasibility study were the high response rate, high completion rate, a group representative of people with MND in demographic and health status characteristics, the use of MND-specific cognitive and health status measures, and the measure used to assess acceptability being nearly identical to the one used in the dignity therapy international randomised controlled trial, which allows for comparison. The limitations include inadequate power to discover small effects, mild to moderate levels of distress at baseline, the lack of a control group, and the use of outcome measures not developed or validated for use with people who have MND. The study group may not be representative of the MND population as a whole as those who selected to participate may have been more likely to think dignity therapy would be beneficial.

**Table 4.** Results of the Participant Feedback Questionnaire Compared to Dignity Therapy and Standard Care in the IRCT [19].

| | People w/MND (*n* = 28) | Dignity therapy IRCT (*n* = 108) | Standard palliative care IRCT (*n* = 111) |
|---|---|---|---|
| **DT has been helpful to me** | 4.18 (0.72) | 4.23 (0.64) | 3.50 (1.01) |
| **DT has been as helpful as any other aspect of my health care** | 3.50 (0.88) | 3.63 (1.04) | 3.27 (1.04) |
| **DT has improved my quality of life** | 3.39 (0.79) | 3.54 (0.95) | 2.96 (0.96) |
| **DT has given me a sense of looking after unfinished business** | 3.68 (0.61) | 3.35 (1.01) | 2.86 (1.60) |
| **DT has improved my spiritual wellbeing** | 3.36 (0.68) | 3.27 (1.09) | 3.00 (1.11) |
| **DT has lessened my sadness or depression** | 3.04 (0.96) | 3.11 (1.02) | 2.57 (0.92) |
| **DT has lessened my sense of feeling a burden to others** | 2.96 (0.92) | 2.81 (0.98) | 2.58 (0.95) |
| **DT has made me feel more worthwhile or valued** | 3.50 (0.79) | 3.38 (0.93) | 3.35 (1.00) |
| **DT has made me feel like I am still me** | 3.71 (0.85) | 3.81 (0.85) | 3.59 (0.92) |
| **DT has given me a greater sense of having control over my life** | 3.18 (0.77) | 3.02 (1.02) | 3.16 (1.00) |
| **DT has helped me to accept the way things are** | 3.54 (0.92) | 3.39 (1.062) | 3.31 (1.01) |
| **DT has made me feel more respected and understood by others** | 3.33 (0.98) | 3.16 (0.90) | 3.04 (0.98) |
| **DT has made me feel that I am still able to carry out important tasks or fill an important role** | 3.61 (0.99) | 3.62 (0.97) | 3.48 (1.00) |
| **I have found DT to be satisfactory** | 4.21 (0.69) | 4.26 (0.63) | 3.80 (0.74) |
| **DT has made me feel that my life currently is more meaningful** | 3.54 (0.69) | 3.55 (1.05) | 3.19 (1.70) |
| **DT has given me a heightened sense of purpose** | 3.32 (0.82) | 3.49 (1.04) | 3.20 (0.98) |
| **DT has given me a heightened sense of dignity** | 3.36 (0.87) | 3.52 (1.04) | 3.09 (1.02) |
| **DT has made me feel more hopeful** | 3.00 (0.86) | N/R | N/R |
| **DT has lessened my suffering** | 3.25 (0.75) | 2.86 (1.04) | 2.70 (1.02) |
| **DT has increased my will to live** | 2.96 (0.98) | 2.94 (1.11) | 2.76 (1.04) |
| **DT has helped me feel closer to people who mean the most to me** | 3.63 (0.97) | N/R | N/R |
| **DT has or will be of help to my family** | 4.00 (0.78) | 3.93 (0.80) | 3.20 (1.00) |
| **DT could change the way my family sees or appreciates me** | 3.48 (1.05) | 3.58 (1.01) | 2.85 (1.00) |
| **I would recommend DT to other patients and family dealing with motor neurone disease** | 4.04 (0.98) | N/R | N/R |

Note: Data are mean (SD). Score 1 is strongly disagree, 2 disagree, 3 neither agree nor disagree, 4 agree, 5 strongly agree. N/R = not reported.

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### Implications for future research

This feasibility study sets the stage for a phase II randomised controlled trial. Potential effectiveness should be further explored through research with people with MND with elevated distress. Research into conducting the intervention via email and through videoconferencing is also indicated. There has been one small study with eight participants showing dignity therapy can be delivered using videoconferencing [41] but a larger study is warranted. Future studies should include hope as an outcome as well as explore the possible relationship between a person's spirituality and changes to hopefulness through dignity therapy.

## Conclusions

Dignity therapy for people with MND is feasible and the unique features of MND, including speech impairment and mild to moderate cognitive impairment can be managed, but the intervention is likely to take a greater length of time to complete compared to previous studies, especially with those individuals experiencing speech impairment who do not utilize assisted communication. Dignity therapy is acceptable to people with MND, who report numerous benefits. Further research is warranted to explore its ability to diminish distress.

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## Author Contributions

Conceived and designed the experiments: BB MO. Performed the experiments: BB. Analyzed the data: BB RK. Wrote the paper: BB MO LB RK.

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# CHAPTER SIX
## 6. Study findings relating to MND family carers

Chapter Six consists of a published manuscript of the study findings relevant to people with MND. The paper is entitled, "Feasibility, acceptability, and potential effectiveness of dignity therapy for family carers of people with motor neurone disease." This paper was published in the open access, peer-reviewed journal *BMC Palliative Care*. The results showed no significant changes on the outcome measures on the group level, but there were some decreases in anxiety and depression in data analyzed on the individual level suggesting dignity therapy may moderate or reduce anxiety and depression in distressed family carers. Family carers saw benefits from dignity therapy to people with MND and to themselves after bereavement, but acceptability was mixed at the time of the intervention with some indicating negative experiences from the therapy. Dignity therapy involving family carers is feasible and involvement of family carers had minimal impact on the therapy.

> One of the worst things about motor neurone disease is watching your loved one slowly fading away day by day, while becoming more and more dependent on their carer. (Sackett and Sakel 2011 p. 2)

Sentiments expressed by MND family carer Brian Sackett in a reflection he wrote about the journey of MND.

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RESEARCH ARTICLE Open Access

# Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neurone disease

Brenda Bentley*, Moira O'Connor, Lauren J Breen and Robert Kane

## Abstract

**Background:** Dignity therapy is a brief psychotherapy that has been shown to enhance the end of life experience. Dignity therapy often involves family carers to support patients weakened by illness and family carers are also the usual recipients of the legacy documents created. No research to date has examined the impact of dignity therapy on family carers at the time of the intervention. This study examined the effects of dignity therapy on family carers of people with motor neurone disease (MND).

**Methods:** This is a cross-sectional study utilizing a one-group pre-test post-test design with 18 family carers of people diagnosed with MND. Outcomes measured caregiver burden, anxiety, depression, and hopefulness. Acceptability was measured with a questionnaire. Feasibility was assessed by examining family carers' involvement in the therapy sessions, time taken to conduct sessions, and any special accommodations or deviations from the dignity therapy protocol.

**Results:** There were no significant pre-test post-test changes on the group level, but there were decreases in anxiety and depression on the individual level. Baseline measures indicate that 50% of family carers had moderate to severe scores for anxiety prior to dignity therapy. MND family carers saw benefits to the person with MND and to themselves after bereavement, but acceptability of dignity therapy at the time of the intervention was mixed with some family carers indicating it was helpful, some indicating it was harmful, and many expressing ambivalence. Dignity therapy involving MND family carers is feasible and the involvement of family carers has minimal impact on the therapy.

**Conclusion:** Dignity therapy is not likely to alleviate caregiver burden in MND family carers, but it may have the ability to decrease or moderate anxiety and depression in distressed MND family carers. Dignity therapy is feasible and generally acceptable to MND family carers. Dignity therapists may provide a better experience for family carers when they are aware of acceptance levels and the quality of partner relationships.

**Trial registration:** ANZCTR Trial Number: ACTRN12611000410954

**Keywords:** Motor neurone disease, Amyotrophic lateral sclerosis, Palliative care, Family carers, Dignity therapy, Psychotherapy, Family therapy

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## Background

Dignity therapy is a brief psychotherapeutic intervention which has been shown to enhance the end of life experience in people with life-limiting conditions [1]. In a distressed group of terminally ill patients, dignity therapy has been shown to reduce anxiety and depression [2]. Dignity therapy is based on a theoretical model of dignity in the terminally ill [3] and its purpose is to reduce dignity-related distress while enhancing hope and meaning [4]. Dignity therapy offers people who are facing death an opportunity to create a legacy document. In a recorded interview, guided by a therapist, a person is invited to recount aspects of his/her life to be remembered, discover purpose and meaning in life, and express final words or advice to loved ones. The therapist and client work together to edit the interview transcript, and the therapy concludes when a final document is produced which can be shared with family and friends [5].

Dignity therapy often involves family carers who may provide support during the therapy interview (s) and editing process, and who may also assist a family member weakened by illness in providing the narrative [6]. Family members are also the usual recipients of the documents, making them an important part of the therapy even when they are not involved in the document's creation. Despite the importance of family members' involvement in dignity therapy, no studies to date have examined the impact of the therapy on family members at the time of the intervention, though dignity therapy has been shown in prior research to moderate the bereavement experience of family members after their loved one had died [7].

This study examined the effects of dignity therapy on the family carers of people with motor neurone disease (MND). A person with MND endures progressive paralysis and gradually loses the abilities to move, speak, swallow and breathe. There is no treatment or cure. Median life expectancy is 2–4 years after diagnosis and death is most often caused by respiratory failure [8]. Family members caring for people with MND often encounter exceptional strain during the caregiving experience due to the rapid and progressive nature of MND coupled with the incapacitating effects of the disease. Research has documented the distress and burden experienced by MND family carers [9-12], and shown the close correlation of distress levels in patient-carer couples [13,14]. Moreover, carer burden increases in MND family carers over time as patient function declines [15-17].

Quality of life may be increased and perceived carer burden decreased in MND family carers who find positive meaning [13,14] and have a sense of hope [18].

Dignity therapy is an intervention designed to bolster hope and meaning and could therefore alleviate perceived burden. Dignity therapy also shows promise to enhance the end of life for people with MND, but its impact on carer distress and burden should be considered when evaluating its overall impact.

### Aims and objectives

The aim of this study was to assess the feasibility, acceptability, and potential effectiveness of dignity therapy for family carers of people with MND. The specific objectives were to assess the impact of dignity therapy on family carers at the time of the intervention by determining whether:

a) dignity therapy decreases perceived caregiver burden, anxiety, depression, and increases hope in MND family carers;
b) dignity therapy is acceptable to MND family carers; and
c) it is feasible to involve MND family carers in the delivery of dignity therapy.

## Methods

### Study design

This cross-sectional study utilized a single treatment group and a pre-test post-test design. A control group was not utilized due to 1) the need to test the feasibility of dignity therapy with people with MND because they are a new research population for this intervention [19]; 2) access issues to the small MND population in Western Australia; and 3) ethical concerns over making a potentially useful intervention unavailable to a control group. More information about the design is available in the study protocol [20].

### Ethical approval

This study was approved by the Curtin University Human Research Ethics Committee (19/2011).

### Setting

MND family carers were recruited from Western Australia (WA) as a result of outreach from the Motor Neurone Disease Association of WA (MNDAWA). MNDAWA provides services to 100–120 diagnosed people with MND at any one time.

### Participants

Family carers of adults diagnosed with MND who could communicate in English were eligible for the study. Enrolment occurred between June 2011 and July 2013. Family carers were invited to participate in the study when a person with MND enrolled to complete dignity therapy as a part of a larger research study. If the family carer did not elect to participate, or if the participant did not have a family carer, the person with MND remained eligible to continue with the study. People with MND

and their family carers were excluded if the person with MND could not provide informed consent (based on the ALS-Cognitive Behavioural Screen [21] and/or the Blessed Orientation Memory Concentration Test (BOMC) [22]), were too ill to complete dignity therapy, or were unable to communicate in English. There was no selection criteria based on distress levels, stage of disease or proximity to death.

### The intervention and study procedures

The intervention was administered by the first author as part of her PhD studies, who was trained in dignity therapy by Harvey Max Chochinov who developed dignity therapy [1,4]. The therapy interviews were audio-recorded and transcribed verbatim. Consistent with the dignity therapy protocol, some family carers provided support and assisted with the interview at the request of the person with MND [5]. The researcher shaped the transcribed interviews using the prescribed editing process [5]. The legacy document was finalised with the aid of the person with MND and, where relevant, his or her family carer. The researcher read the document to each person with MND and to family members who wished to attend the reading. Post-testing occurred via mail or a visit from a second researcher to mitigate response bias. To assure adherence to the dignity therapy protocol, the researcher engaged in regular supervision from Prof. Chochinov and three experienced researchers (two trained in dignity therapy) reviewed three recordings, transcripts and completed documents (10%), which were deemed to be adherent.

### Measures and outcomes

#### *Effectiveness*

Outcome data to measure potential effectiveness were collected from family carers at baseline and one week after completion of dignity therapy. The primary outcome is the family carers' sense of perceived **burden,** measured by the Zarit Burden Inventory [23], a reliable ($\alpha = 0.87$) validated instrument with a summative score ranging from 0–48 where higher scores indicate more burden. Secondary outcomes were: 1) **hopefulness** assessed with the Herth Hope Index [24,25], a reliable ($\alpha = 0.97$) validated instrument developed for people confronting terminal illness and their families with a score ranging from 12–48 and where higher scores indicate more hopefulness; 2) **anxiety** and 3) **depression,** measured with the Hospital Anxiety and Depression Scale (HADS) [26], an instrument often used with family caregivers showing strong reliability ($\alpha = 0.89$) and validity. HADS scores range from 0–21 with scores of 8–10 indicating moderate distress and 11–21 indicating severe distress on both the anxiety and depression subscales.

#### *Acceptability*

A family feedback questionnaire was used to collect family carers' opinions and experiences of the intervention. The questionnaire contained 20 questions answered with a 5-point Likert scale and space for brief explanation.

#### *Feasibility*

Data were collected about the family carers' involvement in the therapy sessions, time taken to conduct the dignity therapy sessions, any special accommodations made in the delivery of the intervention when family carers were involved, deviations from the dignity therapy protocol, reasons for non-completion and reasons for attrition.

#### *Demographic and health status*

Level of impairment of the person with MND and change in physical function over time was collected from the family carer using the Amyotrophic Lateral Sclerosis Functional Rating Scale-R (ALS-FRS) where scores range from 0–48 (lower scores indicating more impairment) [27,28]. Possible cognitive behavioural impairment of the person with MND was assessed with the ALS-CBS, which contains a questionnaire for the person with MND and a separate questionnaire for the family carer [21]. Demographic data on age, gender, relationship to the person with MND, children in the home, caring hours per day, support received, employment status, spirituality, and health history were also collected.

### Analysis

Data were analysed with generalized mixed models (GLMM) as implemented through SPSS's (Version 20) GENLINMIXED procedure. Model parameters were estimated with robust standard errors to accommodate potential violations of the model assumptions. Participant was treated as a random effect and Time (pre-test, post-test) was treated as a fixed effect. Caregiver age, gender, level of education, employment status, spirituality, relationship length, and caring hours were treated as fixed effects and analysed individually as potential moderators of the intervention effect. In order to optimise the likelihood of convergence, a separate GLMM analysis was run for each of the four outcome measures. The GLMM maximum likelihood procedure is a full information estimation procedure that uses all data present at each assessment point, which reduced sampling bias associated with participant attrition. GPower (Version 3.1) indicated that 18 participants would be sufficient to capture relatively 'large' ($f = .36$) pre-post changes on the outcome variables. Descriptive statistics were used to summarize demographic variables and feedback responses.

## Results

### Response rate

We recruited 18 family carers from the study group of 29 people with MND. Six people with MND had family carers who were unwilling or unable to participate (family carer response rate 75%). The reasons for non-participation were either the person with MND did not wish to ask their partner to participate ($n = 3$), or the family carer declined stating they did not have the time ($n = 3$). Five people with MND had no family carers. All 18 family carers completed the study, though one returned only the feedback questionnaire and not the post-test measures.

### Demographic information

Family carers (13 women, 5 men) were aged from 38 to 80 years with a median age of 61. All 18 were spouses/partners who resided with the person with MND. See Table 1 for more demographic information on our study group.

### Baseline levels of impairment and distress for clients and carers

People with MND who were cared for by family carers had low to moderate physical impairment (mean ALS-FRS score was 32.61). Seven carers cared for family members who were mildly to moderately cognitively impaired as measured by the ALS-CBS. The carers reported moderate baseline levels of distress. Half of the family carers had moderate ($n = 6$) to severe ($n = 3$) scores for anxiety. Depression was less common with three carers reporting moderate scores for depression.

### Effectiveness

Family carers reported a significant pre-post increase in burden in conjunction with a significant pre-post decrease in the physical functioning of the patient. After controlling for the pre-post decrease in physical functioning of the person with MND, the pre-post increase in the carer burden was no longer significant ($F$ [1,32] = 3.32, $p = .078$, $d = .30$). There were no significant pre-post changes in self-reported hopefulness, anxiety, or depression. Pre-test and post-test descriptive statistics and test statistics for pre-post differences are reported for all outcome variables in Table 2.

Potential moderators of the intervention effect (caregiver age, gender, level of education, employment status, spiritual beliefs, how long carers had known the patient, caring hours per day, and the number of children living at home) were individually entered in the regression model in order to determine whether positive pre-post changes would emerge at certain levels of the moderator. There was no significant Moderator x Time interactions for any outcomes (all $p$s > .1).

**Table 1 Demographic characteristics of the study group**

| Characteristic | *N* |
|---|---|
| **Gender** | |
| Male | 5 |
| Female | 13 |
| **Age** | |
| 30-39 | 2 |
| 40-49 | 2 |
| 50-59 | 3 |
| 60-69 | 7 |
| 70-79 | 3 |
| 80-89 | 1 |
| **Relationship to person with MND** | |
| Spouse/partner | 18 |
| **Length of relationship to person with MND** | |
| 5 to 10 years | 1 |
| 10 to 25 years | 3 |
| More than 25 years | 14 |
| **Residence area** | |
| Urban/metropolitan | 14 |
| Rural | 4 |
| **Highest level of education attained** | |
| Primary/elementary school | 1 |
| Secondary/high school | 11 |
| University/technical | 5 |
| Postgraduate | 1 |
| **Number of minor children living at home** | |
| 0 | 15 |
| 1 | 2 |
| 3 | 1 |
| **Current employment status** | |
| None | 13 |
| Full-time | 3 |
| Part-time | 2 |
| **Caring hours per day** | |
| Less than 4 hours | 5 |
| 4 to 8 hours | 2 |
| 8 to 12 hours | 1 |
| More than 12 hours | 10 |
| **Time since MND diagnosis of family member** | |
| Less than one year | 4 |
| One to two years | 9 |
| Two to three years | 2 |
| Three to four years | 0 |
| More than four years | 3 |

**Table 1 Demographic characteristics of the study group** *(Continued)*

| | |
|---|---|
| **Are you currently being treated for a major medical condition?** | |
| No | 6 |
| Yes | 12 |
| **Have you been prescribed medication to help you cope?** | |
| Anti-depressant | 1 |
| Anti-anxiety | 1 |
| No | 16 |
| **Do you consider yourself to be a spiritual person?** | |
| Yes | 3 |
| Somewhat | 10 |
| No | 5 |
| **Cognitive screening scores of person with MND** | |
| No impairment | 11 |
| Suspected mild to moderate impairment | 7 |
| **Formal support received to help cope with family member's MND[a]** | |
| None | 7 |
| Support group | 3 |
| Home care | 4 |
| Respite | 1 |
| Counselling | 1 |
| Psychologist/Psychiatrist | 2 |
| Other: | |
| MND Association | 3 |
| Church | 1 |
| Massage | 1 |
| Medication | 1 |

*Note.* [a]*Participants could list more than one type of support.*

A reliable change (RC) score for each carer [29] was computed to investigate the presence of reliable pre-post change at the individual rather than group level. The RC score is the degree to which the person changes on the outcome variable divided by the standard error of difference between the pre- and post-test scores. When the absolute value of the RC score is greater than 1.96, (Wise [30] has argued that this value can be reduced in some situations), it is likely that the post-test score reflects a *real* or *reliable* change. The results suggest potential prevention effects for anxiety and depression, and both increases and decreases in hopefulness (see Table 3).

**Table 2 Mean pre-test post-test scores (and standard deviations) for measures of burden, hopefulness, anxiety, depression, and physical function**

| Outcome | Pre-test $N = 18$ | Post-test $N = 17$ | $F$ (1.33) | $P$ | $d$ |
|---|---|---|---|---|---|
| Caregiver burden (ZBI) | 12.44 (7.89) | 16.29(11.22) | 5.58 | .024 | 0.95 |
| Hopefulness (HHI) | 38.39 (4.46) | 36.71 (4.52) | 3.19 | .083 | 0.62 |
| Anxiety (HADS) | 7.28 (3.71) | 6.88 (4.33) | 1.33 | .257 | 0.26 |
| Depression (HADS) | 4.17 (3.33) | 4.41 (3.91) | 0.03 | .860 | 0.39 |
| Physical function (ALS-FRS) | 32.61 (9.76) | 30.12 (9.62) | 7.00 | .012 | 1.19 |

### Acceptability

The reported benefits and acceptability of dignity therapy to the family carers of people with MND was mixed. There were many feedback responses indicating it was helpful, some indicating it was not helpful or even harmful, and some showing ambivalence. Half ($n = 9$) of the family carers agreed or strongly agreed that dignity therapy was helpful to them, while about a quarter ($n = 4$) disagreed or strongly disagreed. More family carers disagreed than agreed that dignity therapy helped to reduce their feelings of stress as a carer ($n = 5$ v $n = 6$) and an equal numbers agreed as disagreed that it helped them feel closer to their partner ($n = 6$ v $n = 6$). Some family carers appeared to find dignity therapy confronting and mentioned the attention it brought to their partners' impending death.

Family carers were more positive about seeing benefits to their partner, with 16 agreeing or strongly agreeing that dignity therapy had been helpful to their family member, 11 reporting it was an important component of their care, 10 reporting it had increased meaning in their partner's life, and 9 indicating it had helped their family member prepare for the end of life. Despite mixed feelings about the therapy, 13 believe the document would be an ongoing source of comfort and 14 would recommend dignity therapy to other people with MND and their families. The complete results of the feedback questionnaire are reported in Table 4. Selected comments are available in Table 5.

### Feasibility

Twelve of the 18 family carers assisted with the interview and editing process. Ten family carers provided support by attending the therapy sessions and contributing to the narrative when requested by their partner, one served as a proxy for her husband who had lost the ability to speak and write (completing the entire interview

**Table 3 Number of carers showing reliable improvement, deterioration, and no change for burden, hopefulness, anxiety, and depression**

| Outcome | Improved | Deteriorated | No change | $N$ |
|---|---|---|---|---|
| Caregiver burden (ZBI) | 0 | 4 | 13 | 17 |
| Hopefulness (HHI) | 3 | 8 | 6 | 17 |
| Anxiety (HADS) | 2 | 0 | 15 | 17 |
| Depression (HADS) | 1 | 1 | 15 | 17 |

**Table 4 Results of the Family Feedback Questionnaire**

| Item | Family carers ($N = 18$) | | | |
|---|---|---|---|---|
| | Mean | SD | *n* Agree (SA + A) | *n* Disagree (SD + D) |
| **DT was helpful to my family member** | 4.22 | 0.647 | 16 | 0 |
| **DT has given my family member a heightened sense of purpose or meaning** | 3.87 | 1.060 | 10 | 2 |
| **DT helped increase my family member's sense of dignity** | 3.56 | 0.984 | 8 | 2 |
| **DT helped prepare my family member for the end of life, whenever that may occur** | 3.33 | 0.970 | 9 | 3 |
| **DT was an important component of my family member's care as any other aspect of their care, including symptom management** | 3.61 | 0.979 | 11 | 3 |
| **DT helped reduce my family member's suffering** | 3.22 | 1.003 | 7 | 5 |
| **DT helped increase my family member's hopefulness about the future** | 3.17 | 0.857 | 6 | 4 |
| **The DT document helped me during this time of our life** | 3.33 | 1.085 | 9 | 4 |
| **DT has helped me prepare for the end of life of my family member, whenever that may occur** | 3.11 | 0.832 | 5 | 4 |
| **DT was helpful in reducing my feelings of stress as a carer** | 3.00 | 0.907 | 5 | 6 |
| **DT helped me feel closer to my family member** | 2.94 | 0.938 | 6 | 6 |
| **DT has increased my hopefulness about the future** | 3.11 | 0.758 | 6 | 4 |
| **The DT document will continue to be a source of comfort for my family and me** | 3.83 | 0.618 | 13 | 0 |
| **I would recommend DT to other patients of family members who are dealing with MND** | 4.00 | 0.686 | 14 | 0 |
| **Scoring: 1 *strongly disagree*, 2 *disagree*, 3 *neither agree or disagree*, 4 *agree*, 5 *strongly agree*.** | | | | |

**Table 5 Selected comments from the family feedback questionnaire**

| Item | Comment |
|---|---|
| The dignity therapy document helped me during this time of our life. | "It has provided a source of information and inspiration". |
| | "I didn't learn anything about him I didn't already know". |
| | "I put my husband first and yet what I read I didn't feel very appreciated or loved". |
| Dignity therapy was helpful in reducing my feelings of stress as a carer. | "Answering the questions actually increases the stress". |
| | "When the real suffering begins the stress is going to come no matter what". |
| | "[There is] more understanding, less tension". |
| Dignity therapy helped me feel closer to my family member. | "Some days we are on the same page, but other days we are upset, angry and not close at all". |
| | "Nothing replaces 50+ years of constant close companionship and mutual caring". |
| | "We have always been close but I feel more protective now". |
| Dignity therapy has helped me prepare for the end of life of my family member, whenever that may occur. | "I don't see how relating his life in a few short pages could prepare me". |
| | "One thing it did do was focus on the end and not to live and enjoy the journey along the way the best we can". |
| | "[It helped] from pushing aside the situation to more acceptance". |
| Dignity therapy was helpful to my family member. | "Just for him to think of the past and what he has achieved in his life is satisfying". |
| | "She expressed emotions which she normally suppresses". |
| | "He enjoyed the opportunity to put memories on paper and have something concrete for others to read". |
| Dignity therapy helped prepare my family member for the end of life, whenever that may occur. | "[We] recently went on a family holiday and he was able to talk to his children about his condition". |
| | "He's been in denial but has recently come to terms with his diagnosis and the dignity therapy helped through giving an opportunity to talk about these issues". |
| | "It has made him face up to his situation and to express himself to family and friends". |
| The dignity therapy document will continue to be a source of comfort for my family and me. | "If we are missing him, we can just read the booklet". |
| | "The document will provide a basis for reference and reflection". |
| | "We will treasure his story forever". |

in his presence with minimal contribution from her husband), and one carer of a speech impaired wife was significantly involved in providing detail and elaborating on her responses which were written on a whiteboard. However, not all speech impaired participants required or requested assistance from their family carers as several relied on assisted communication, including email, to complete the interview and editing. We analysed the data using descriptive statistics to determine if the distress levels or acceptability levels differed between the family carers involved and those not involved in the interviews and editing and we found no significant differences. The family member who served as a proxy was the only deviation from the dignity therapy protocol, though Chochinov and colleagues report instances of dignity therapy conducted via family proxies and it appears that this is an acceptable deviation [6].

Generally, individual dignity therapy meetings involving family carers ($n$ = 12) were of a longer duration than those completed by the client alone ($n$ = 17). Family carers added to the session dialog, asked questions, and often provided refreshments or engaged in other caring tasks that made their partner more comfortable. All of these actions extended sessions. Nonetheless, involvement of family carers did not equate to longer legacy documents. For clients assisted by family carers, documents were 7 to 47 pages (mean 20.42, *SD* 13.35), while documents were 11 to 57 pages (mean 22.94, *SD* 10.615) for clients who completed the therapy alone. The number of sessions required to complete the therapy was fewer in the group assisted by family carers (mean 3.75 v 4.41), and the days to complete the intervention slightly more in the group assisted by family carers (mean 46 v 39).

## Discussion

Dignity therapy has been shown to moderate the bereavement experience in family carers when they were interviewed 9 to 12 months after death [7], but no previous studies have looked at the impact of the therapy on family carers at the time of the intervention. We hypothesised post-intervention decreases in burden, anxiety and depression scores and an increase in hope, but there were no significant changes. Rather, our population showed an increase in burden which correlated to a decline in the physical function of the patient during the study period. This effect is consistent with research of burden over time in MND family carers [15-17]. Without a control group, we are unable to ascertain whether dignity therapy had a prevention effect against expected increases in burden, anxiety, and depression, or a decline in hopefulness.

The individual results on anxiety and depression are more encouraging and suggest that dignity therapy has the potential to decrease anxiety and depression in family carers who are experiencing moderate to high levels of distress. This is similar to the findings for terminally ill cancer patients, where distressed individuals had a decrease in anxiety and depression scores [2], but those with low baseline levels of distress showed no change [1].

Acceptability of dignity therapy was mixed. Family carers felt that the therapy provided a benefit to their family members and that the document would help them in bereavement, and most rated the experience as satisfactory and one they would recommend to others. Whether a family carer was directly involved in the therapy had little impact on the acceptability or feasibility of dignity therapy. Rather, the comments provided on the feedback questionnaire suggest that family carers' level of acceptance of their partner's imminent death, or the quality of the relationship between family carer and partner, may lead to dignity therapy having a potentially negative impact on family carers at the time of the intervention.

### Strengths and limitations

The strengths of this study were the high response rate and high completion rate of MND family carers, the use of MND-specific cognitive and health status measures, and the demographic characteristics of the sample are generally representative of MND family carers. The limitations include inadequate power to discover small to moderate effects, mild to moderate levels of distress at baseline, and the lack of a control group. The study group may not be representative of all MND family carers because those who declined to participate may have been more distressed, and people with MND with severe cognitive impairment and their family carers were excluded from the study.

### Implications for future research

The effectiveness of dignity therapy in decreasing perceived caregiver burden, anxiety or depression, or increasing hopefulness in MND family carers could not be determined in this study. A randomised controlled trial with a greater number of participants is needed, perhaps incorporating a stepped-wedge or cluster design. An experimental study focusing on distressed family carers is warranted to determine if dignity therapy has the potential to decrease anxiety and depression. A qualitative study with family carers is indicated to explore more fully the mixed acceptability results provided in the feedback questionnaire. Further, a longitudinal study would determine if the document was helpful to carers following bereavement.

### Ethical challenges

Dignity therapy may trigger emotional upset in people with MND who are experiencing emotional lability and this was frequently encountered. Emotional lability, also

known as pseudobulbar affect, is an MND symptom which has the potential to cause distress to the person with MND and their family carer if it is not treated sensitively and therapeutically. In our study, we provided a supportive and safe environment, information and psycho-education, and normalised the symptom. Other ethical challenges encountered include negotiating complex relationships of participants and family carers, minimising harm to both parties as well as extended family members through what was written in (or left out of) the generativity document, and balancing the interests of the family carers with those of the patient. These challenges suggest that the skillful application of dignity therapy by a trained psychotherapist who is knowledgeable about MND is paramount in any future research.

## Conclusions

The findings of this study suggest that dignity therapy is not likely to alleviate the burden encountered by MND family carers during caring, but it may have the ability to decrease or moderate anxiety and depression in distressed MND family carers. Dignity therapy is feasible and generally acceptable to family carers of people with MND, who recognize benefits to their ill family member and also the potential benefits to themselves during bereavement as a result of having the legacy document. Comments from carers indicate that it is important for a dignity therapist to be aware of acceptance levels and dynamics in partner relationships in order to best provide a satisfactory experience for family carers.

#### Competing interest

The authors declare that they have no competing interests.

#### Authors' contributions

BB and MO designed the study. MO and LB supervised the research. BB conducted the research and drafted the article. RK provided substantial assistance with the data analysis. MO, LB, and RK made substantial contributions to the critical revision of the article. All authors read and approved the final manuscript.

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# CHAPTER SEVEN
## 7. Feasibility and Implementation Issues

Chapter Seven expands on issues related to the feasibility of dignity therapy with people who have MND that were introduced in Chapters Five and Six. It explores the ways common MND symptoms discussed in Chapter Two including speech loss and impairment, emotional lability, cognitive impairment and paralysis can affect the provision of dignity therapy and psychotherapy more generally. This chapter also provides a detailed account of the practical aspects of performing dignity therapy with people with MND, including the time and costs involved and characteristics of completed documents, and adds to the body of literature addressing the delivery of dignity therapy in a clinical setting first introduced in Chapter Three. A personal reflection published in the *Journal of Palliative Medicine* entitled "It Takes the Time that it Takes" is incorporated to provide insight into time delays and endorsement from dignity therapy's creator for acceptance of these delays in certain cases. Ethical considerations relating to the delivery of dignity therapy with people with speech or cognitive impairment are discussed. Future directions for research relating to the feasibility and implementation of dignity therapy as part of standard palliative psychosocial care are discussed.

> I'm not sure how much more of this disease I can take. The deterioration of my tongue and throat muscles is making it harder for me to speak. I think as clearly as I ever have, but I just can't mouth my words clearly any more. Even though it saddens me beyond belief, I can't cry, because when I cry I can't breathe. When I can't breathe I panic because I feel I am closer to death. (Finlay 2008 p. 128).

Maree Finlay of Western Australia was diagnosed with MND at age 37 in 2004. During her illness, she wrote a memoir documenting her early life and her journey with MND. The above passage was written shortly before her death in December 2007.

## 7.1 Introduction

As established in Chapter Two, MND is a terminal disease of short prognosis, and management of MND focuses on controlling symptoms and maintaining QOL from diagnosis until death (Simmons 2005, McDermott and Shaw 2008). A review of the literature in Chapter Three suggests that providing psychosocial support to both people with MND and their family carers is critical to maintaining QOL (Ganzini, Johnston et al. 1999, Simmons, Bremer et al. 2000, Clarke, Hickey et al. 2001, Cupp, Simmons et al. 2011); yet only a small percentage of MND research is dedicated to the psychosocial aspects of MND and intervention studies are rare (Pagnini, Simmons et al. 2012, Aoun, Bentley et al. 2013). Moreover, there is no research dedicated to the practical aspects of delivering psychosocial care to people with MND, a group that has challenging physical symptoms including cognitive-behavioral impairment, speech loss or speech impairment, emotional lability, and paralysis. In order to provide effective psychosocial care in palliative care settings, an exploration of how common MND symptoms can affect the efficacy and feasibility of psychotherapeutic interventions is needed (Kasl-Godley, King et al. 2014). This study of dignity therapy with people with MND and their family carers provides an opportunity for such an examination.

## 7.2 Background

As described in Chapter Three, dignity therapy is an end-of-life psychosocial intervention that was developed and tested for a palliative care population where 95% to 97% have malignant conditions (Chochinov, Hack et al. 2005, Chochinov, Kristjanson et al. 2011). The results of prior dignity therapy research with cancer patients is not directly transferable to people with MND as demographic features, baselines levels of distress, ability to communicate, physical impairment, and cognitive acuity are some of the factors that differ between these groups and may impact on the delivery of the intervention. A close examination of feasibility issues is necessary (Craig, Dieppe et al. 2008), which will inform the feasibility of dignity therapy with people with MND specifically, and also the use of psychosocial and psychotherapeutic interventions with people with MND and their families more generally.

Previous research examining practical details about the delivery of dignity therapy, including the required clinical time and costs, the length of time needed to complete dignity therapy from start to finish, and the length of dignity therapy documents, has reported incomplete and inconsistent results (See Chapter 3, section 3.4.4.8). A detailed appraisal of practical aspects of implementing dignity therapy gleaned from the present study will provide information about delivering psychosocial interventions to people with MND, and add to the knowledge base on the time and resources needed to implement dignity therapy into clinical practice.

The aim of this chapter is to explore further feasibility issues related to conducting a psychotherapeutic intervention with people who have MND and their family carers, to present data on the practical aspects of dignity therapy with this group, and to add to the knowledge base on the time and resources needed to implement dignity therapy into clinical practice. In Chapter Five, it was reported that the increased time required to deliver dignity therapy to some people with MND may be prohibitive. Key challenges include: the increased time required to deliver the intervention to people who may have impaired speech and time to travel to a palliative care group who are primarily cared for at home. However, common symptoms like mild to moderate cognitive impairment, speech impairment and emotional lability did not detract from the perceived benefits or acceptability of the therapy based on the outcome measures (p. 85-86). In Chapter Six, it was reported that it is feasible to involve MND family carers in dignity therapy even though the meeting times were of a longer duration in this group than in people with MND who did not involve family in the therapy sessions. There did not appear to be any differences in acceptability levels between family carers who were involved in the therapy and those who were not involved based on responses in the feedback questionnaire (p. 93-94). This chapter expands upon these findings by providing a detailed report and analysis of the feasibility issues involved in providing dignity therapy to people with MND, which will inform practice guidelines for psychotherapists working with this group.

## 7.3 Methods

### 7.3.1 Study design

This cross-sectional study utilized a one-group pre-test post-test design with 29 people with MND and 18 MND family carers. (For more information on the methodology, see Chapters 4-6).

### 7.3.2 Measures and outcomes

#### 7.3.2.1 Demographic and health status

Demographic data including age, gender, location, and marital status were collected. Disease specific health related QOL was measured with the ALSAQ-5, where scores range from 0-20 and higher scores denote decreased health related QOL (Jenkinson, Fitzpatrick et al. 2007). Probable FTD/cognitive behavioral impairment was screened with the ALS-CBS with scores ranging from 0-20 and lower scores indicating impairment (Woolley, York et al. 2010).

#### 7.3.2.2 Contact sheet

A contact sheet was used to record data at each participant meeting, including the number of dignity therapy sessions and the duration of each session. Details about final documents produced in dignity therapy were recorded, such as the number of copies requested, the length of documents, and any materials added to documents. The contact sheet also contained space for the researcher to write brief notes related to the impact of MND symptoms on the delivery of the intervention, including observations and impressions of physical symptoms, non-verbal behaviors, challenges encountered, and notable comments from participants and family carers.

#### 7.3.2.3 Audio recordings and transcription cost

The total duration of audio recordings per participant was determined and the cost of transcriptions was calculated. Most audio recordings were transcribed by a professional transcriber at a cost of $1.80 AUD per recorded minute which is the usual rate in WA. In some recordings, where speech impairment made the recordings very difficult to understand, the transcriptions were completed by the researcher at no cost. However, the overall cost of transcriptions has been calculated by noting the minutes per

recording and multiplying them by the transcription rate per minute for all recordings, including those completed by the researcher.

### 7.3.3 Analysis

Descriptive statistics were used to summarize the variables using SPSS version 20.

## 7.4 Results

### 7.4.1 Demographic information

Participants with MND were 20 men and 9 women who ranged in age from 32 to 81. MND family carers who participated were 13 women and 5 men who ranged in age from 38 to 80. Participants resided throughout WA where the one-way travel distance ranged from 0 kilometers (km) for one participant who preferred to work at the researcher's office and another who completed the intervention via Skype, to 430 km with a median of 24 km. In all instances, both people with MND and MND family carers engaged in the research from the same location (See Table 7-1 for more demographic information).

### 7.4.2 Impairment

Twenty-four participants had low to moderate health-related QOL according to the ALSAQ-5, and 13 had mild to moderate suspected cognitive impairment according to scores on the ALS-CBS. Five symptoms that affected the delivery of dignity therapy were noted on the contact sheets and include: emotional lability, speech loss, speech impairment, cognitive impairment, and paralysis. Over 80% ($N$=24) of participants displayed at least one of these symptoms during the intervention. Often more than one symptom was present in the same individual. Seven participants, or about one-quarter, had multiple symptoms that were consistent with bulbar presentation of MND, where upper motor neurons are affected and speech loss or impairment, cognitive impairment and/or emotional lability occur early in the disease. (See Table 7-2 for more information on ALSAQ-5 scores, ALS-CBS scores, and symptoms observed during dignity therapy, which together create impairment profiles for each participant with MND).

**Table 7-1 Participant Demographic Information**

| NO. | AGE | MARRIED? | SPOUSE PARTICIPATED? | ONE-WAY TRAVEL (KM) |
|---|---|---|---|---|
| **1** | 64 | YES | YES | 22 |
| **2** | 65 | YES | YES | 430 |
| **3** | 65 | YES | YES | 352 |
| **4** | 75 | YES | YES | 17 |
| **5** | 32 | YES | | 5 |
| **6** | 63 | YES | YES | 17 |
| **7** | 63 | | | 8 |
| **8** | 71 | | | 28 |
| **9** | 80 | YES | YES | 38 |
| **10** | 55 | YES | YES | 164 |
| **11** | 68 | YES | YES | 300 |
| **12** | 58 | YES | YES | 6 |
| **13** | 42 | YES | YES | 0 |
| **14** | 69 | YES | | 319 |
| **15** | 54 | YES | YES | 169 |
| **16** | 52 | YES | YES | 18 |
| **17** | 76 | YES | | 22 |
| **18** | 77 | | | 24 |
| **19** | 71 | YES | | 3 |
| **20** | 64 | YES | YES | 17 |
| **21** | 81 | | | 50 |
| **22** | 69 | YES | YES | 17 |
| **23** | 69 | YES | YES | 33 |
| **24** | 68 | YES | | 93 |
| **25** | 63 | YES | YES | 300 |
| **26** | 60 | YES | | 0 |
| **27** | 62 | YES | YES | 30 |
| **28** | 64 | | | 21 |
| **29** | 72 | YES | YES | 164 |

#### 7.4.2.1 Emotional lability

The most commonly observed symptom that had an impact on the intervention was emotional lability. This was observed in 9 participants - 6 men and 3 women - who experienced periods of uncontrollable crying during dignity therapy. A few participants, both people with MND and their family carers, knew this type of crying was a symptom of MND but other participants were surprised when the symptom occurred. For example, in one observation of emotional lability the researcher noted, “X said he has a problem with emotional lability and reports that sometimes he starts crying and can’t stop,” and in another noted, “X found she was crying while telling parts of her childhood story and didn’t understand why she was so emotional when she didn’t feel sad.” One

participant, who experienced uncontrollable crying during the completion of the pre-test questionnaires and expressed embarrassment, discontinued involvement with the study explaining that the process was "stirring things up that I don't want stirred up." One participant, who was ventilator dependent, had irregular breath patterns and was gasping while crying, which was startling for the researcher, though based on the behavior of family members present during data collection, these symptoms seemed routine for the family.

**Table 7-2 Impairment profile of participants with MND**

| No. | Health-related QOL* | Suspected cognitive impairment** | Observed cognitive impairment | Observed speech impairment | Observed speech loss | Observed emotional lability | Observed paralysis of hands |
|---|---|---|---|---|---|---|---|
| 1 | Moderate | Mild | YES | | | | |
| 2 | Moderate | Moderate | YES | | | | |
| 3 | High | Normal | | | | | YES |
| 4 | Moderate | Normal | | mild | | YES | |
| 5 | Low | Normal | | | | | YES |
| 6 | Moderate | Normal | | | YES | | |
| 7 | High | Normal | | | | | YES |
| 8 | Low | Normal | | | YES | | |
| 9 | Moderate | Moderate | | | YES | | |
| 10 | Moderate | Mild | YES | mild | | YES | |
| 11 | Moderate | Normal | | | | | |
| 12 | Low | Normal | | YES | | YES | |
| 13 | High | Normal | | | | | YES |
| 14 | Moderate | Normal | | | | YES | |
| 15 | Moderate | Moderate | YES | | YES | YES | |
| 16 | Moderate | Moderate | | YES | | | |
| 17 | High | Normal | | | | | |
| 18 | High | Moderate | | | | | |
| 19 | Moderate | Moderate | | | | | YES |
| 20 | Moderate | Normal | | | | YES | |
| 21 | Moderate | Mild | YES | | | | |
| 22 | High | Normal | | | | | |
| 23 | Moderate | Moderate | | | YES | YES | |
| 24 | Moderate | Normal | | mild | | | |
| 25 | Low | Moderate | | | YES | YES | |
| 26 | Moderate | Normal | | YES | | | |
| 27 | Moderate | Mild | | mild | | YES | |
| 28 | High | Mild | | | | | |
| 29 | Low | Normal | | mild | | | |

*ALSAQ-5 score = 1-6 high, 7-13 mod, 14-20 low

**ALS-CBS score = 17-20 normal, 13-16 mild, 10-12 moderate

Though numbers are small and only tentative conclusions can be reached, women with MND did not seem to be as disturbed by emotional lability as men with MND. This may be due to the influence of masculine norms where crying is less socially acceptable for men than for women (Mahalik, Locke et al. 2003). On the whole, men with MND expressed embarrassment and often appeared to avoid emotionally evocative areas during the dignity therapy interview, focusing more on biographical information and accomplishments, and less on messages of love and hope.

When emotional lability was encountered, the Protocol for Minimizing Risk of Emotional and Psychological Harm (see Appendix X) was activated and participants were offered continued emotional support through MNDAWA. No participants expressed that they were distressed by dignity therapy or symptoms of emotional lability, and none sought continued support.

#### 7.4.2.2 Speech loss

Six participants had lost the ability to communicate verbally and relied on AAC to complete dignity therapy. Two, who primarily used tablet devices to communicate, completed the intervention entirely by email. After the initial meeting, dignity therapy questions were asked and answered via email in one or two interactions per week. These interactions were compiled and edited in a similar fashion to the oral interview transcripts, creating a comprehensive document identical to the other dignity therapy transcripts. One participant completed the process via email with ease but the second participant became discouraged after a few interactions and commented “there’s not a lot of plot to this novel.” A visit was arranged to read aloud to the participant the document that had been produced. The participant was pleased with the meeting and thereafter completed the interview through email with enthusiasm.

The third participant with speech loss completed the intervention using a Lightwriter, a portable type-input voice out-put device. This participant was a retired secretary who easily adapted to typing on the device which would then speak aloud what she had written in an electronic voice. The interview was completed in two sessions which were audio recorded and transcribed following the usual protocol. Another participant who had only recently begun using the Lightwriter, and was having difficulty

adapting to using it, used a combination of the Lightwriter, handwritten notes, gestures and verbalizations. This interview was audio recorded and the handwritten notes were read aloud by the researcher, along with the meaning of gestures and verbalizations. This recording was not coherent and difficult to transcribe. The participant also wrote out some notes between interview sessions which were added to the document.

A fifth participant with speech loss used a whiteboard to write cues and then had her spouse provide details for the narrative. This participant also experienced emotional lability and her cognitive screen score suggested moderate impairment. This was evidenced by nonsensical phrases (aphasia) on the whiteboard but these were understood by her spouse. The sixth participant was ventilator dependent and could not speak. This participant relied exclusively on his spouse to tell his life story, though he guided his wife's storytelling with hand gestures. On a few occasions when meaning could not be deciphered from the gestures, the participant wrote a brief note, which took considerable time and effort.

#### 7.4.2.3 Speech impairment

Eight people had impaired speech. Of these, five exhibited mild impairment which resulted in occasions where words or phrases were not immediately understood, but overall this level of impairment had minimal impact on the intervention. In 3 people, the impairment was considerable and the participants did not use AAC, which made comprehension difficult. Interviews were performed and audio recordings were created, though the transcripts were difficult and extremely time consuming to create as there were many unintelligible words, sentences or paragraphs that had to be revisited in subsequent meetings. In each of these cases, the researcher/interviewer performed the transcription as the recordings were too difficult for the transcriptionist to understand. This also created the opportunity for the researcher to re-listen to interviews during which time the researcher noticed that there was a substantial amount of content that was not understood and not responded to during the interview. For example, one participant revealed childhood abuse and another said he had disgraced his family name. There was no response from the researcher to either of these comments as the researcher had not understood what was said at the time (though the comments were addressed in

subsequent meetings, and the participants were provided information and referral for psychological support, which they refused). The three participants with significant speech impairment expressed frustration but only with themselves, and they did not become disillusioned or stop dignity therapy. Ultimately, each of these participants requested to do some of their own editing via email. These three participants had the longest completion times and some of the longest documents.

#### 7.4.2.4 Cognitive impairment

Cognitive impairment was observed in 5 participants whose ALS-CBS scores indicated mild to moderate impairment. One participant, whose screen suggested mild FTD, had short and long term memory impairment. The participant's spouse was visibly anxious and distressed by the participant's memory loss during the dignity therapy interview. After the spouse tried different methods to prompt memories, the issue was discussed and a decision was made for the spouse to leave the session and go for a drive in order to avoid further distress. This participant created one of the shortest documents, though the spouse was very happy and comforted by the document, stating "It really helped me – I read it all the time. When I'm worried about what I will do in the future, I read the booklet."

A second participant with a score approaching the cut-off on the cognitive screening measure appeared to have behavioral variant FTD. While this participant reported he was employed in a position of authority from time-to-time and managed the family finances, there appeared to be impairment in his executive functioning. This participant made inappropriate, insensitive and sometimes bizarre comments during dignity therapy, including some that were hurtful to his spouse. This participant's transcript was edited and reduced more than any other to remove nonsensical or objectionable content. In the end, the participant was pleased with dignity therapy, who said it was "of the upmost importance" while the spouse was not, stating, "Hearing him put me down to someone else ... only increased my stress."

The cognitive impairment observed in the remaining 3 participants, such as memory lapses or aphasia, had minimal impact on the intervention. It should be noted

that 8 other participants had scores suggesting cognitive impairment on the ALS-CBS but impairment was not observed during the intervention.

#### 7.4.2.5 Paralysis

Six participants had paralysis that led to an inability to type or grip a writing instrument. As a result, these participants could not hold or write on their documents, correspond with the researcher or use non-voice activated AAC methods. In all instances, this symptom had minimal impact on the intervention and the necessary adjustments were easily made.

**Table 7-3 Time and costs to perform dignity therapy**

| | *Minimum** | *Maximum* | *Median* | *Mean* | *Std. Deviation* |
|---|---|---|---|---|---|
| ***Total Visits*** | 3 | 7 | 4 | 4.14 | 10.94 |
| ***Total Contact Hours*** | 3.25 | 11.25 | 5.75 | 5.82 | 1.87 |
| ***Total Days*** | 7 | 152 | 26 | 42 | 36.25 |
| ***Recorded Minutes*** | 31 | 254 | 91 | 98.48 | 50.76 |
| ***Transcription Cost*** | $55.80 | $457.20 | $163.80 | $177.27 | $91.37 |

* Note: Email clients with no recorded interview time were excluded in the above figures on recorded minutes and transcription cost. The median and range are reported as the data are not normally distributed, and mean and SD are also included for use in comparison with other studies.

### 7.4.3 Time and costs associated with dignity therapy

Total intervention visits (excluding post-testing) ranged from 3 to 7 visits with a median of 4. Hours of clinician/participant contact time ranged from 3.25 to 11.25 with a median of 5.75 hours. This time includes obtaining consent and the completion of pre-test questionnaires in the initial meeting where dignity therapy was explained (see p. 53) but does not include the completion of post-test questionnaires as this occurred after dignity therapy had concluded and was collected by a second researcher or by mail to reduce response bias. Days from the initial visit to providing the final document ranged from 7 days to 152 days with a median of 26. Recorded interview minutes ranged from 0 (with two participants who were unable to speak and who completed dignity therapy exclusively via email) to 254 (median of 81). When the two participants with no recordings were excluded, the range was 31 to 254 minutes with a median of 91 minutes

per recording. The median cost per recording for all participants was $145.80. When the two participants with no transcriptions were excluded, the range was $55.80 to $457.20 with a median of $163.80. (Note: this method does not take into account the actual time spent transcribing which could be considerably longer in those with speech impairment; See Table 7-3).

### 7.4.4 Characteristics of documents

Participants requested from 1 to 20 copies of their finished document with a median of 4. Documents ranged from 2,025 words to 11,978 words with a median of 6,003. Upon request to the researcher, participants were able to add material to their documents prior to completion. Ten participants requested to add material, most commonly a photograph on the cover page and/or a collection of photographs added as an appendix. Other materials appended to documents included poetry and songs authored by two participants, a timeline of important dates, a record showing dates of postings on naval vessels, and two participants included letters of commendation from previous employers. Nineteen participants did not request the addition of materials, but 5 of these requested an electronic copy of their document and said they or a family member would add material such as photographs in the future. Children of two of these participants began adding photographs before the final documents were delivered in hard copy form (see Table 7-4).

**Table 7-4 Final document characteristics**

| | *Minimum* | *Maximum* | *Median* | *Mean* | *Std. Deviation* |
|---|---|---|---|---|---|
| ***# of Copies Requested*** | 1 | 20 | 4 | 5.21 | 3.77 |
| ***Page Length*** | 7 | 57 | 20 | 21.90 | 11.66 |
| ***Word Length*** | 2025 | 11978 | 6003 | 6202.1 | 2989.09 |

* Note: The median and range are reported as the data are not normally distributed, and mean and SD are also included for use in comparison with other studies.

## 7.5 Discussion

### 7.5.1. Effect of physical impairment and symptoms on dignity therapy

While people with MND and their families are often advised to seek support from mental health professionals to help cope with MND (MND Association 2014, State Government of Victoria 2014), and research articles addressing QOL and psychosocial distress in MND frequently recommend increased psychosocial support for people with MND and their family carers (Mitsumoto, Bromberg et al. 2005, McLeod and Clarke 2007, Aoun, Bentley et al. 2013), there is a paucity of evidence available to practitioners about the practical aspects of providing psychosocial support, including how common symptoms and physical impairments encountered in people with MND may affect counseling and the efficacy of interventions. In this study, the majority of participants had MND impairments which had at least some effect on the delivery of dignity therapy and participation in psychotherapy more generally. In most instances, the presence of MND symptoms posed minor challenges during the delivery of the intervention. Paralysis, minor speech impairment, and speech loss where participants used AAC had little effect on dignity therapy as long as allowances were made, such as a slower pace of conversation, to accommodate the symptoms and enhance communication between the researcher and participant.

Mild to moderate cognitive impairment was not an issue in most cases, though there were two exceptions. In the first, an MND family carer became distressed by the observed memory loss of her spouse and his difficulty with telling his story. Even though this MND participant had passed a cognitive screen, cognitive decline has been shown to have a profound negative impact on MND family carers, with increases in burden and depression and decreases in QOL (Chio, Vignola et al. 2010). Relieving the family carer of her responsibilities of care during the intervention allowed dignity therapy to be completed to the satisfaction of both parties; however, Chochinov warns that performing the intervention with people who have cognitive impairment has the potential for harm if it results in a dignity therapy document that is a distorted representation of the person (Chochinov 2012). In the second case, the participant with MND was suspected of behavioral variant FTD based on his score on a cognitive screen and observed behavior, which included impulsivity, self-centeredness and lack of empathy, although the participant and family carer did not appear to recognize the presence of cognitive impairment. Research has found that people with MND with FTD

lack insight of the magnitude of their behavioral change and suggests that MND family carers may fail to recognize neurobehavioral symptoms that develop slowly (Woolley, Moore et al. 2010). Though this participant found dignity therapy to be acceptable and reported benefits, the difficulty of performing the intervention, along with the distress caused to the MND family carer as a result of hearing her husband criticize her, together indicate dignity therapy may not be suitable for people suspected to have behavioral variant FTD. Increased education and screening is indicated to improve awareness about this symptom and to provide better care in the multidisciplinary setting (Wicks and Frost 2008).

Emotional lability was encountered most often and had the potential to cause distress for both the participant and family carer. Not only can emotional lability be distressing for people with MND (Moore, Gresham et al. 1997, Mitchell and Borasio 2007), but previous research has found a relationship between psychological distress in MND family carers and demonstrations of emotional lability by care recipients (Goldstein, Atkins et al. 2006). Often, participants and MND family carers in this study were unaware that the behavior was a symptom of MND, a finding that is consistent with previous research (Elman, Houghton et al. 2007). Chochinov states that dignity therapy "is often quite emotional; even emotionally neutral material can resonate in ways that are quite poignant" (Chochinov and McKeen 2011 p. 87). While information exists on the medical management of emotional lability for people with MND and other neurological disorders (Meininger 2005, Ahmed and Simmons 2013), no guidance can be found on performing psychotherapy or psychological interventions with people who have this symptom. Drawing on counseling theories and practices (Kleinke 2002, Corey 2005), when emotional lability occurred, the researcher created a safe and supportive environment by providing information and education about the symptom, and normalizing the occurrences. However, the efficacy of psychotherapeutic interventions is presumably affected in instances where clients are unable or unwilling to discuss emotionally evocative topics through fear of triggering a potentially embarrassing and distressing symptom such as emotional lability (Moore, Gresham et al. 1997). Increased education about emotional lability is needed for health care providers, people with MND, and family members to increase awareness about this symptom (Goldstein,

Atkins et al. 2006, Wicks and Frost 2008, Ahmed and Simmons 2013). The use of screening measures may help identify people with MND who are struggling with this symptom and improve care (Moore, Gresham et al. 1997). A possible solution for people distressed by symptoms of emotional lability is providing the option of online therapy, as research shows that some people prefer online therapy due to the reduced emotional exposure experienced through the use of this method (King, Bambling et al. 2006).

Significant speech impairment where participants did not use AAC had the most effect on the intervention and on the delivery of psychotherapy more generally. Verbatim transcripts of interviews were very difficult to create and more visits were required over a longer period of time to obtain and clarify the narrative. Research examining unaided communication between a person with significant speech impairment and a speech therapist demonstrates not only the difficulty of communication but also the intense effort that must be expended by both parties in the dyad for a negligible level of comprehension to occur (Damico, Simmons-Mackie et al. 2006). Research addressing the communication effectiveness of people with MND has determined that communication is difficult in all situations when intelligibility falls below 70% on a communication measure (Ball, Beukelman et al. 2004). Previous research addressing the provision of psychotherapy to people with speech loss and impairment suggests that AAC devices are a necessity, with one author stating "The devices do not merely facilitate the therapeutic process; they enable it to occur" (Crawford 1987 p. 97). Similarly, Chochinov affirms, "Conditions that interfere with a patient's ability to communicate can be challenging and, in some instances, may preclude their being able to participate" (Chochinov 2012 p. 56). He also states that the participant, therapist and transcriptionist "must all speak the same language" (Chochinov 2012 p. 58). Chochinov does not elaborate or provide strategies for a dignity therapist to assess an acceptable level of comprehension, but he implies understanding between the parties is a paramount concern.

In this study, the amount of important content that was not heard or comprehended while performing dignity therapy with people who had significant speech impairment, including instances of painful self-disclosure, undermined the creation of a

therapeutic relationship and environment. A central principle of person-centered psychotherapy is that a therapist must demonstrate empathic understanding of their clients. Corey (2005) states, "One of the main tasks of the therapist is to understand clients' experience and feelings sensitively and accurately as they are revealed in the moment-to-moment interaction during the therapy session" (p. 173) and Chochinov confirms, "therapists must pay careful attention to everything that is happening between them, including what is said, how it is said …" during dignity therapy (Chochinov 2012 p. 77). In participants with significant speech impairment, the ability of the researcher to understand and respond to content was diminished. These findings suggest that dignity therapy with people with significant speech impairment, for example those scoring below a determined cut-off point on the Modified Communication Effectiveness Index (CETI-m) (Ball, Beukelman et al. 2004) or the Assessment of Intelligibility of Dysarthric Speech (AIDS) (Yorkston, Beukelman et al. 1984), may best be accomplished through an alternate method rather than a verbal interview, such as through the use of AAC or email.

While dignity therapy took the longest to perform with the three people who had significant speech impairment in this study, this is not a sufficient reason to discourage the use of the intervention with this group. Chochinov states, "Conditions that interfere with a patient's ability to communicate can be challenging" and "creative thinking, patience and ingenuity" should be used in these cases (Chochinov 2012 p. 56). Research with people living with dysarthria has documented the psychological distress, including threats to self-image and isolation, that occur when barriers to societal participation are encountered due to their disability (Walshe and Miller 2011), and speech impairment adversely affects the QOL of people with MND and their family members (Murphy 2004). Research on the interactions between health care providers and people with speech impairment has shown that patients are controlled and often feel judged during interactions, and that a culture of partnership is needed to provide meaningful interventions (Gordon, Ellis-Hill et al. 2009, Walshe and Miller 2011). Based on the needs of this group, a longer completion time is of little importance. The researcher also consulted with Professor Chochinov who created dignity therapy about the long completion time with one of the speech impaired participants. The personal reflection at

the end of this chapter, “It Takes the Time That It Takes” was written and published as a result. In summary, Professor Chochinov highlighted the necessity of psychosocial care and suggested that the length of time taken to complete the intervention was less important than the potential benefits experienced as a result of dignity therapy. The experience detailed in the reflection supports this position.

Participants with speech loss who used AAC were able to complete dignity therapy and they reported comparable benefits to those who could speak. The most efficient and novel AAC method was the use of email. The use of email to perform psychotherapy is new and evolving with themes about its clinical use both suggesting optimism about reaching new client populations balanced by caution over the significant changes to traditional counseling methods brought about by technology (Goss and Anthony 2009). As in this research, the use of technology in psychotherapy is often guided by client needs rather than by practitioners (Goss and Anthony 2009). While one participant using email to complete dignity therapy did become discouraged by the relative isolation of this method, a face-to-face interaction restored the enthusiasm and resolve of the participant. The resulting positive experience of both people who had lost the ability to speak and who completed dignity therapy by email indicates that dignity therapy may be successful even in the absence of a personal therapeutic relationship. Averill et al (2013) found that people with MND who had difficulty expressing themselves experienced increased psychological wellbeing as a result of a written expressive disclosure intervention where people with MND wrote or spoke into a tape recorder about their deepest thoughts and feelings for 20 minutes a day on three occasions over the course of a week. Dignity therapy may present a similar opportunity for those who have lost the ability to speak to benefit from personal written expression.

### 7.5.2 Feasibility and implementation

In this study, dignity therapy averaged 4 sessions with client contact time of 6 hours. This is similar to the studies by Hall (2012) with 29 aged care residents and Montross (2011) with 23 cancer patients. Clinician editing time completed out of the presence of the client was not tracked with every participant in this research, but where tracked it ranged from 4 to 8 hours of clinician editing time per recorded hour of

interview. An estimate of the average editing time performed outside of the presence of the participant in this study was 9 hours per participant, which is only slightly above the 8.69 hours reported by Hall (2012), and both this research and Hall's report a similar average of approximately 15 hours of clinician time per participant. The mean total recorded interview time per participant was 1 hour 38 minutes in this study, which is comparable to the 1 hour 36 minutes reported by Hall (2012), but more than the 66 minutes reported by Johns (2013) with 4 cancer patients. The mean duration of the therapy from start to finish was 42 days, while Hall (2012) reported 31.81 days.

Despite similarities between this study and the Montross (2011) study in number of sessions and client contact time, the documents in the present research were considerably longer. In the Montross study, the mean number of words per document was 2,993, while documents in this study were 6,202 words long. (Documents averaged 5,878 in Johns' (2013) study while Hall (2012) did not report on the length of dignity therapy documents). The mean cost of transcription in the Montross study was $56 USD (Montross, Winters et al. 2011), while it was $165 AUD here. Accounting for currency exchanges, the transcription cost for the present study in Australia was about three times more expensive than in the US; however, it was less than the "$200 CAD" per transcript that Chochinov suggests as a budget (Chochinov 2012 p. 176). While inconsistencies and incomplete data continue to exist, the picture is becoming clearer on the time commitment and other practical aspects required to perform dignity therapy.

While the cost of transcription was detailed in two studies, previous studies have not tracked other costs such as travel costs, the cost of paper, copies and binding supplies, phone, mail and internet costs, and equipment costs for audio recorders, computers and printers, (which was also the case in the present study). Moreover, while recent research is providing clarity concerning the number of visits and time required to meet with clients and edit documents, travel time, time spent arranging visits, waiting time, and cancelled appointments have not been considered fully in this or other research, although Hall (2012) noted these were common issues that increased the time commitment required.

Delivering dignity therapy to rural and remote participants in this study was particularly time intensive. Dignity therapy with four participants, who all resided over 300 km from the Perth metropolitan area, required considerable travel time and at least one night's accommodation. In addition, precision planning and around the clock work by both the transcriptionist and researcher were needed to accomplish parts of the intervention during each two-day/one-night period, which may not be feasible in all settings. An additional 3 participants resided over 150 km from Perth and required a full day for each client visit when travel was considered. However, it should be noted that challenges such as these which result from the cost of travelling across vast geographical distances are not unique to dignity therapy.

The use of e-health methods, including videoconferencing and email, present a time and cost-efficient method to deliver dignity therapy to rural and remote locations. In addition to the two participants who completed dignity therapy via email, one participant who resided on the opposite side of Australia successfully completed dignity therapy using videoconferencing and reported numerous benefits. This supports previous research by Passik and colleagues (2004) that dignity therapy can be effectively delivered using e-health methods. In addition, research has shown that psychological interventions using e-health technologies are just as effective as face-to-face therapeutic interventions for people who are unable to access 'traditional' forms of care (Barak, Hen et al. 2008), although users require training to use these technologies competently and ethically (Shandley, Klein et al. 2011).

### 7.5.3 Implications for future research

Future research detailing the time required to complete dignity therapy is indicated, along with a cost analysis exploring its use in inpatient, outpatient and rural/remote settings. Larger studies of delivering dignity therapy using e-health, including email and videoconferencing, are also indicated to overcome speech loss and impairment, provide an optional environment of reduced emotional exposure for those experiencing emotional lability, and to deal with the expense and difficulties encountered in serving rural and remote populations.

An additional avenue of research concerns appropriate cognitive screening tools and cut-off scores for potential recipients of dignity therapy, with an emphasis on detecting behavioral variant FTD. Chochinov states "One of the most important reasons to exclude patients from dignity therapy is impaired cognitive ability, limiting the patient's capacity to providing meaningful and reflective responses (Chochinov 2012 p. 60); however, there is little guidance on appropriate screening tools or cut-off scores to exclude participants from receiving dignity therapy. Several dignity therapy studies, including the IRCT, used the Blessed Orientation Memory Concentration (BOMC) test (Katzman, Brown et al. 1983) to exclude participants with dementia, although brief measures like the BOMC to assess cognitive ability have been shown to be unreliable in screening for FTD in people with MND (Floris, Borghero et al. 2012).

### 7.5.4 Ethical challenges

None of the participants in this study had been screened or tested for cognitive impairment previously, and no participant disclosed to the researcher that cognitive impairment had been discussed with them by their health care providers. In addition, several participants were not informed that emotional lability was a physical symptom of MND. These factors presented an ethical challenge to the researcher, who was in the position of disclosing information about physical symptoms to patients outside the care received through their multi-disciplinary care team. When this occurred, the researcher encouraged participants and family carers to discuss these symptoms with their health care providers. When given permission by the participant, the researcher also attempted to liaise with one or more health care providers.

Performing psychotherapy in an environment where a considerable amount of the content is not understood by the therapist creates an ethical challenge relating to therapist competence and the potential for harm. Corey states, "Competence is required of practitioners if they are to avoid doing harm to their clients" (Corey, Schneider Corey et al. 2007 p. 314). Performing dignity therapy with someone who has severe speech impairment but does not use AAC is akin to performing psychotherapy with a client who speaks in a language the therapist barely understands, a task described by a psychiatrist writing to the Lancet in 1963 as "well-nigh impossible" (Enoch 1963 p. 1163) and one

that is contrary to Chochinov's (2012) inclusion criteria that the therapist, transcriptionist and participant are fluent in the same language. The potential for harm is compounded in instances where disclosures of trauma, such as childhood sexual abuse, are missed and as a result ignored during the therapeutic interaction, as occurred in this research. Numerous studies have found that negative responses from those receiving a traumatic disclosure, such as minimizing the event or an absence of a supportive response, can result in harm to the client (Ullman 1996, Andrews, Brewin et al. 2003, Becker-Blease and Freyd 2006).

## 7.6 Conclusion

This chapter explored the ways in which common MND symptoms affect dignity therapy, and indicates that behavioral variant FTD, emotional lability and significant speech impairment are issues that need to be further addressed. It examined the time and resources necessary to deliver dignity therapy with people who have MND and compared this evidence with information from other studies. The results of these investigations suggest that psychosocial care providers for people with MND should be educated about common symptoms and the disease trajectory. Screenings may help identify people with MND with cognitive behavioral impairment, emotional lability, and speech impairment, which will enable psychotherapists to engage with participants and their families in optimal ways. E-health methods, including e-mail and videoconferencing, may provide satisfactory alternatives for providing dignity therapy to people with speech impairment and emotional lability. The use of e-health methods may also address concerns about the cost-effectiveness of dignity therapy, which research suggests is more time intensive and expensive than originally reported, as well as provide a viable option for serving those in rural and remote areas who may not otherwise have access to this form of support.

**7.7 Bentley, B. (2012). "It takes the time that it takes." Journal of Palliative Medicine 15(8)**

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Personal Reflection

# It Takes the Time That It Takes

Brenda Bentley, M.A., M.P.A., MFTi

IT WAS FIVE DAYS before my first Christmas in Perth, Australia, and our family had begun to arrive the night before. As the rest of household slept, I quickly threw on a skirt and tank top and left to attend the memorial service for one of my study participants, a 57-year old Anglican minister who had died from motor neurone disease (MND). Though I feared I would be late, I could see people milling about in the shaded church courtyard as I pulled up and parked. Walking in, I didn't know anyone except Stuart's wife Grace and the minister, who had stopped in one day for coffee as we had been doing our work creating Stuart's Dignity Therapy transcript.

Because Dignity Therapy involves the creation of a manuscript about important memories and messages that are to be left behind after death, I had heard a lot about Stuart's three children and their families, his parents and in-laws, his brothers and sister, friends, and other important people who appeared in the narrative of Stuart's life, but I felt a stranger at this gathering and headed quickly to the empty seats at the back of the church. Grace came from nowhere and intercepted me in the middle of the aisle, giving me a long hug. Letting her tears flow, she said, "Thank you so much for being here. It means so much to me and it would mean so much to Stuart." I opened my mouth and nothing came out. I stammered under the weight of the loss, the memories, Grace's gratitude, and my inexperience. This was the first memorial service I attended where my relationship was professional, and I had hoped I would have something more comforting and eloquent to offer than a gaping mouth but I didn't, and it was okay.

I had first met Stuart Jordan in July. He had telephoned the day before I left for vacation and, sounding as though his tongue was three times too big for his mouth, asked if he and his wife could take part in my PhD research on Dignity Therapy with people who have motor neurone disease. I explained that I was going on vacation the next day, and we made an appointment for two days after my return. I hastily sent a confirmation letter to "Stuart Jones," as I had misunderstood his name to be. Within hours, I was on a plane to visit family and friends in California, a place I had left just nine months earlier to pursue this research and a place where motor neuron disease is known as amyotrophic lateral sclerosis or Lou Gehrig's disease.

My first day back in Perth, jet-lagged and dressed in jeans and a t-shirt, I received a call from Stuart's wife. "Where are you?" she asked. "We have an appointment." I pulled up the confirmation letter on my computer and saw I had written the wrong date. I also noticed a glaring typo in my congratulations regarding the news of their first grandchild. "So many mistakes. How are they ever going to put their trust in me? " I thought to myself. "I made a mistake with my calendar, but can I come now?'' I asked Grace apologetically.

Our first meeting lasted over an hour. Their small house located across from a train station was neat and tidy. Jolly, the dog, greeted me at the door. A stereo and shelves housing a large record collection dominated the small living room. Grace offered me a cappuccino from an impressive steaming machine in the kitchen, and Stuart had an espresso. Stuart was firmly in the grip of MND. He could not use his left hand. Eating, standing, and walking were an extraordinary effort. He slurred his words and it was very difficult to understand him, but he easily forgave my mistakes and imperfections and opened himself up. When I asked him to rate as a problem "Feeling like I am no longer who I was," he burst out crying.

Stuart and I met six more times over the next ten weeks and also emailed his document back and forth between us. During this time his health quickly declined. He had his PEG feeding tube inserted. He and Grace went on a trip, which turned out to be a disaster when he was ill the entire time and required several emergency room admissions. He began using a VPAP breathing machine to improve his oxygen levels. He was twice admitted and then released from the palliative care unit at a local hospital. As time went on, I began to wonder if telling his story was helping to keep him alive. Each time I thought we were done, he wanted to include more text or more photos. When I thought we couldn't possibly add any more, he came up with the idea for a "timeline," which meant we had more work to do.

While working with Stuart, I often wondered if I was doing Dignity Therapy wrong. These thoughts did not occur to me as often with other participants, who more easily stayed within the flexible boundaries of the study protocol and the Dignity Therapy manual. Mostly, I was concerned because I was spending more time with Stuart than I had with other study participants, and it was taking longer than the others to finish. When I asked the creator of Dignity Therapy, Dr. Harvey Max Chochinov, about my concern in one of our supervisory phone calls, he acknowledged it was taking longer than usual, but stressed the importance of providing psychosocial support to people with terminal illness. Finally, in

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his sage and experienced way, he made my concerns seem silly when he said simply, "It takes the time that it takes!"

But I had other concerns as well. Were Stuart and Grace becoming too comfortable with me, and was I letting them overstep therapist/researcher boundaries? During one visit to the palliative care unit to complete the captions on his photographs, Stuart asked for a hug before I left. During a visit at their home, Grace noticed I needed a haircut and the next thing I knew she had dialled a hair salon and was handing me the phone. Even the topics Stuart discussed in our interviews were a challenge for me. The interviews were dominated by Stuart reflecting on his relationship with God, and I often felt out of my depth guiding a deeply religious man through an interview that addressed issues of spirituality at the end of life.

About a month after we had completed our work and the study, Grace requested a visit from me. After a short chat with both of them, Grace, who was under an amazing amount of stress and who was having difficulty coping, asked if I would stay with Stuart while she ran an errand. I sat with Stuart while he dozed. His forehead was pulled tight and he seemed turned inward, as if having to concentrate on each laborious breath. I asked if he wanted to try guided imagery to help him relax and he chose a scenario that took place at a beach. He urinated into his bedside urinal twice and I emptied and cleaned it. After forty minutes, Grace returned and I quietly left. It was the last time I was to see Stuart.

Grace emailed six weeks later "just letting you know Stuart passed away yesterday morning around 8:30 a.m. ...How we love the memories of Stuart that you have left behind for us. Thank you. Love, Grace."

When I reflect on Stuart, I recall my questions and blunders and marvel at how it all added up to a therapeutic relationship at the end of Stuart's life that was supportive and meaningful for Stuart and his family, and a research experience that was rich. For a psychotherapist who works from a person-centered and existential frame of reference, I realize that working with Stuart was one of my best achievements thus far, rather than fraught with error. As Irvin Yalom advises new therapists, "Therapy should not be theory-driven but relationship-driven."[1] My relationship with Grace and Stuart was real, supportive, and empathic. Stuart mattered to me, and I am the better woman for it.

## Acknowledgements

The names of the patient and his wife have been changed to protect their anonymity. The author wishes to thank "Grace," who consented to the publication of this essay. She would also like to thank her academic supervisors, Prof. Samar Aoun, Dr. Moira O'Connor, and Dr. Harvey Max Chochinov for their guidance and support and for reviewing this essay, as well as Gali Hagel, who provided expert writing advice.

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# CHAPTER EIGHT
## 8. Discussion

Chapter Eight concludes this thesis with a discussion of the key findings of this research in relation to people with MND, MND family carers, and dignity therapy. The findings advance the empirical knowledge base in three key areas relevant to palliative care research and practice. These findings highlight recommendations regarding the use of dignity therapy with people with MND and their family carers, including: the use of screening measures for cognitive-behavioral impairment, speech impairment, and emotional lability; choices for people with MND about how to complete dignity therapy, including the use of e-health methods; and caution when dealing with families who have complex histories, conflictual patterns of relating or other dysfunction, with families who are struggling to accept the terminal nature of MND, and with participants and family members from non-Western cultures. It reports the strengths and limitations of the research, and recommendations for future research are outlined in the areas of efficacy, family members, and translation of dignity therapy into practice.

**No One**

Time, passing away
You're here for a day, and then you are gone
No time for a song, listened along, with no-one
I, I could have sailed fast
I should have done more to get us to shore
But that's in the past, I'm tied to a mast, with no-one
What's the point of regret?
You give what you get and get what you give
The life that you live, is yours, it belongs to no-one
You play, you win or you lose,
It's all down to chance, there's nothing to choose
There's nothing to bet, you can't be in debt, to no-one
And now you see, looking at me
Far from the life I could have achieved
I whistle my song, I'm singing along, with no-one, no-one

Time, passing away
You're here for a day, and then you are gone
No time for a song, listened along, with no-one (Banks 2011)

Lyrics from the song "No One" by Alistair Banks. Banks was diagnosed with MND in July 2010 and became known as "Alistair the Optimist" when he was chosen to represent a campaign to raise awareness about MND in the UK. During this time, Banks wrote songs and recorded a CD in fulfilment of a final wish to raise funds for MND research and to leave a legacy for his family and friends. Banks died in January 2013.

## 8.1 Introduction

A diagnosis of MND is a shocking and devastating experience for people with MND and their families (O'Brien, Whitehead et al. 2011a, Mistry and Simpson 2013). People with MND experience unremitting loss during the course of the disease as they gradually lose physical function and abilities (Foley, Timonen et al. 2014). Because there is no curative treatment, care goals for people with MND and their families focus on maintaining QOL (Simmons 2013). QOL for people with MND does not correlate with physical impairment and does not decline over time despite declining abilities (Simmons, Bremer et al. 2000, Robbins, Simmons et al. 2001, Cupp, Simmons et al. 2011). Rather, psychological, psychosocial and existential factors, such as depression, hopefulness, spirituality, family, and relationships, play the most important roles in maintaining QOL (Chio, Gauthier et al. 2004, Roach and Averill 2009, Gibbons, Thornton et al. 2013). Compared to other disease groups, people with MND are at an increased risk of suicide (Fang, Valdimarsdottir et al. 2008) and seek out hastened death more frequently (Ganzini, Silveira et al. 2002) as a result of psychosocial distress and decreased QOL. Like people with MND, QOL in MND family carers is not related to disease severity (Rabkin, Wagner et al. 2000, Murphy, Felgoise et al. 2009) and is instead related to psychological and psychosocial factors (Trail, Nelson et al. 2004, Lo Coco, Lo Coco et al. 2005, Calvo, Moglia et al. 2011). Considering the importance of psychological and psychosocial distress in maintaining QOL in people with MND and their family carers, there is a lack of interventions (Aoun, Bentley et al. 2013) and an absence of research and direction on providing this type of care (Pagnini, Simmons et al. 2012).

The aims of this study were to explore the feasibility, acceptability and potential benefits of dignity therapy for people with MND and their families. Dignity therapy is based on an empirical understanding of dignity-related distress in the terminally ill (Chochinov, Hack et al. 2002b) and has been shown to enhance the end of life experience in people with cancer (Chochinov, Kristjanson et al. 2011). Through an exploration of dignity therapy with people with MND, this study has advanced the knowledge base in three key areas for palliative care today as outlined in Chapter One. These areas include the expansion of end of life care that is historically grounded in cancer care to providing care to all people equitably at the end of life regardless of disease; the provision of support to family carers of the terminally ill who form part of the unit of care according to palliative care definitions; and the provision of psychosocial care to people at the end of life in order to address their 'total pain,' including psychological, psychosocial, and existential dimensions of distress. Specifically, it has investigated the potential of dignity therapy to enhance the end of life experience for people with MND thus extending this supportive end of life intervention to people other than those with cancer, examined the effect of dignity therapy on MND family carers who shoulder substantial burden and experience distress, and advanced research addressing psychosocial care at the end of life through making a novel and significant contribution to dignity therapy research and practice.

This chapter concludes the thesis by presenting key findings in the three key areas outlined in Chapter One: people with MND, MND family carers, and dignity therapy research and practice. It provides recommendations for the clinical use of dignity therapy with people with MND and their families, reports the strengths and limitations of the research, and makes recommendations for future research in the areas of efficacy for people with MND, the effects of dignity therapy on MND family carers, and translation of dignity therapy into practice.

## 8.2 Expansion of end of life care beyond cancer: People with MND

### 8.2.1 Key findings

The findings indicate that people with MND report satisfaction with dignity therapy. Satisfaction with dignity therapy in this study was comparable to previous

dignity therapy studies involving people with cancer (Chochinov, Hack et al. 2005, Chochinov, Kristjanson et al. 2011, Hall, Goddard et al. 2011), and was higher than satisfaction levels reported in a Danish study of people with cancer (Houmann, Chochinov et al. 2014) where all studies used the same measure. Reported benefits in this study include enhanced meaning and purpose, improved family relationships, increased acceptance, strengthened identity, and decreased aftermath concerns. In the context of living with a diagnosis of MND, distress is frequently experienced by people with MND and their family carers and psychological interventions are suggested (McLeod and Clarke 2007, Aoun, Bentley et al. 2013), but there is an absence of theoretically driven, empirically tested psychosocial interventions (Pagnini, Simmons et al. 2012) and an unmet need reported by people with MND and their family carers for emotional, psychosocial and existential support (Foley, O'Mahony et al. 2007, Whitehead, O'Brien et al. 2012). These findings are a promising step to meet the psychological support needs of people with MND and their family carers.

There were no significant pre-test post-test changes on the outcomes measuring hopefulness, spirituality and dignity, most likely due to the small sample size achievable in Western Australia, but there were increases ($N$=4) and decreases ($N$=7) in hopefulness at the individual level. Although numbers are small, there appears to be a relationship between hopefulness and spirituality as all participants with increased hope after dignity therapy reported they were both spiritual and religious on a demographic questionnaire, while almost half of those with decreased hope indicated they were neither spiritual nor religious. A strong relationship between these variables was also reported in other research, including a recent review which found that 29 of 40 studies examining the relationship between hope and spirituality reported a significant positive relationship, while no studies found an inverse relationship (Koenig 2012). There also appears to be a relationship between hope and spirituality in people with MND, and previous research has reported that people with MND who are spiritual are more hopeful (Murphy, Albert et al. 2000). In addition, hope has been described as the key to maintaining QOL in people with MND (Worthington 1996). Dignity therapy's potential to increase hope in people with MND who identify as spiritual warrants further investigation.

In terms of physical impairment, twenty-four participants (over 80%) had at least one physical symptom which affected the delivery of dignity therapy. Physical symptoms observed during dignity therapy included speech loss, speech impairment, emotional lability, cognitive impairment, and paralysis. Seven participants, or about one-quarter, had multiple symptoms. In all seven participants, the multiple symptoms observed were consistent with bulbar presentation of MND, where upper motor neurons are affected and speech loss or impairment, cognitive impairment and/or emotional lability occur early in the disease. Emotional lability was encountered most often during dignity therapy, which required appropriate knowledge, training and support from the therapist in order to manage therapeutically. Paralysis and minor speech impairment had the least impact on the delivery of dignity therapy and allowances were easily made to accommodate these symptoms. In most cases, mild to moderate cognitive impairment did not affect the intervention. However, in one case the difficulty of conducting dignity therapy along with the negative impact on the family carer indicates that the intervention may not be feasible, acceptable and/or beneficial in cases where behavioral variant FTD is suspected. Cognitive and behavioral decline in people with MND relates to increased distress and decreased QOL in MND family carers (Chio, Vignola et al. 2010, Merrilees, Klapper et al. 2010), and behavioral changes have the greatest impact on caregiver burden (Lillo, Mioshi et al. 2012). Providing dignity therapy to people suspected of behavioral variant FTD has the potential to worsen the already high levels of distress and burden reported in MND family carers who care for those with symptoms of FTD (Chio, Vignola et al. 2010, Merrilees, Klapper et al. 2010, Lillo, Mioshi et al. 2012), as those participants may lack the empathy, insight, and deliberation required to complete dignity therapy (Lillo, Mioshi et al. 2011).

The findings relating to the impact of MND symptoms on dignity therapy indicates that it is imperative for clinicians who provide dignity therapy to be knowledgeable about MND symptoms and the disease trajectory, and that they develop the necessary skills and expertise to work with this group. Previous research has shown that people with MND and their families indicate that health care professionals often lack knowledge, skills, and training about MND (Hughes, Sinha et al. 2005, O'Brien, Whitehead et al. 2011b, Foley, Timonen et al. 2012). These are also pertinent concerns

for psychosocial care providers. While there are undoubtedly many counsellors and psychologists currently supporting people with MND, there is an absence of empirical research to guide practice, theoretically grounded interventions, training, and protocols in this area (Haley, Larson et al. 2003, Pagnini, Simmons et al. 2012, Kasl-Godley, King et al. 2014).

Delivery of dignity therapy to people with significant speech impairment who did not use AAC presented challenges that may be insurmountable due to a number of practical and ethical issues, including: the extended length of time needed to complete dignity therapy with these participants; the difficulties encountered preparing transcripts; the resulting increased costs; and the ethical challenges created by the amount of content that was missed, misunderstood or misinterpreted by the researcher/therapist. Participants with speech loss who used AAC required some modification to the dignity therapy protocol to accommodate the use of AAC methods, including performing the intervention through e-mail or through a family member proxy. In previous research, videoconferencing was used to deliver dignity therapy (Passik, Kirsh et al. 2004) and one study allowed participants to help edit their documents via email (Johns 2013), but this study is the first to explore delivery of the intervention using email, including the dignity therapy interview. Family member proxies have been used in previous dignity therapy research where participants had dementia (Chochinov, Cann et al. 2012) but this is the first study to explore the use of a family member proxy due to speech loss and physical impairment. Psychologists in palliative care settings are challenged to evaluate and modify practices to accommodate patient symptoms (Kasl-Godley, King et al. 2014). The findings of this study indicate the use of family member proxies and email are viable methods of delivery for people with MND who have speech loss or impairment.

While dignity therapy was designed for people with advanced cancer in the final weeks of life, dignity therapy may be offered to people with MND any time after diagnosis as people with MND have no hope for a cure. As such, modifications to the delivery of dignity therapy were necessary to accommodate people with MND. The diagnosis of MND is highly distressing (O'Brien, Whitehead et al. 2011a, Mistry and Simpson 2013) and people with MND and their families express the need for greater

psychosocial support at diagnosis (O'Brien, Whitehead et al. 2011a). In this research, one family carer suggested that dignity therapy would be better if provided soon after diagnosis, while others felt it would be better delivered closer to death. Other options for the delivery of dignity therapy with people with MND include using e-health methods to accommodate people located a distance from services who may not otherwise have access to this form of support, with people with speech loss who rely on assistive forms of communication, and with people with emotional lability who may prefer the reduced emotional exposure of this method. Another option for people with MND is allowing them to take the time they need to complete their generativity document to their satisfaction, whether it is a week or even months (Bentley 2012). When there is limited evidence available about the potential efficacy of different psychotherapeutic approaches in palliative care settings among different disease groups, therapists should extrapolate best practices (Kasl-Godley, King et al. 2014), as occurred in this research.

The findings indicate that providing *choices* about whether and how to engage in dignity therapy is important for people with MND. Recent research has found that people with MND have unmet expectations concerning their care (Foley, Timonen et al. 2012) and an expressed need for a broad range of services, including psychosocial care (Whitehead, O'Brien et al. 2012). However, exercising choice about whether and how to participate in an intervention is paramount as people with MND adapt to unremitting losses by exerting control over health care services and by choosing how and when they will engage with services (O'Brien, Whitehead et al. 2011b, Foley, Timonen et al. 2014). A central concern for people with MND is their perception of control over their treatment choices (Hogden, Greenfield et al. 2012).

Based on the findings, dignity therapy may not appeal to people who are distressed, including people with MND who are experiencing hopelessness, loss of meaning, or who request a hastened death. The majority of people who responded to recruitment efforts in this research had low baseline levels of distress. Similarly, participants recruited to other dignity therapy studies also had low baseline levels of distress (Chochinov, Kristjanson et al. 2011, Hall, Goddard et al. 2011, Houmann, Chochinov et al. 2014). There is conflicting research findings on whether dignity

therapy may alleviate distress in people with elevated distress, with one study reporting the potential to alleviate anxiety and depression in cancer patients with high baseline levels of distress (Juliao, Oliveira et al. 2012) and others reporting the complexities of delivering the intervention to distressed people may preclude the use of dignity therapy with this group (Johns 2013, Hall, Goddard et al. 2013c). Previous research has found that dignity therapy is considered "too big" of an undertaking for some patients (Houmann, Chochinov et al. 2014 p. 9), including people who are absorbed with coping with the physical, psychosocial and existential stressors encountered as a result of their condition (Houmann, Rydahl-Hansen et al. 2010, Johns 2013, Hall, Goddard et al. 2013c, Houmann, Chochinov et al. 2014). For example, in Johns' study (2013), three people did not complete dignity therapy stating it required too much energy and time, and they preferred to focus on other things, and Hall concluded "It is difficult to find the essence of patients when they are worried or unhappy" (Hall, Goddard et al. 2013c p. 1751).

Interestingly, it appears some cultural groups find dignity therapy too confronting (Houmann, Rydahl-Hansen et al. 2010, Akechi, Akazawa et al. 2012), and it may conflict with cultural norms such as aversions to self-praise and self-pride (Houmann, Rydahl-Hansen et al. 2010) or to open discussions about death (Akechi, Akazawa et al. 2012). One example from this research was an immigrant from Southeast Asia who engaged in dignity therapy in order to create a document for his Western wife and children, but cautioned that he did not want his document shared with his parents as it could be construed as disrespectful and culturally unacceptable. This area warrants further exploration. As dignity therapy has not been widely tested with non-Western cultural groups, caution and careful consideration is needed when working with these participants.

## 8.3 Providing support to family carers of the terminally ill: MND family carers

### 8.3.1 Key findings

There were no significant pre-test post-test changes on the outcomes measuring caregiver burden, anxiety, depression or hope in the small sample group of 18 MND family carers. There were increases ($N$=3) and decreases ($N$=8) in hopefulness on the

individual level, but unlike the findings in people with MND, we found no relationship between changes in hopefulness and reported spirituality. The MND family carers in this study were moderately distressed according to baseline scores on the HADS, and 3 had decreases in anxiety (*N*=2) and depression (*N*=1) post-intervention according to RC scores. Though the numbers are small, these preliminary results indicate that dignity therapy may have the potential to decrease anxiety and depression in MND family carers. This finding warrants further investigation.

Slightly lower scores were reported on the family feedback questionnaire in this study than the scores reported in previous research. This could be due to data collection times, as in this study responses were collected two weeks after completion of the therapy, while responses were collected 6 to 12 months after death in previous dignity therapy research (McClement, Chochinov et al. 2007). Here, 89% of family carers reported that dignity therapy was helpful to the participant and 72% reported the dignity therapy document would be a continuing source of comfort after bereavement, while the numbers were 95% and 77% respectively in the dignity therapy pilot study with cancer patients (McClement, Chochinov et al. 2007). Previous research has found that MND family carers express a need for emotional support and counselling during their caring experience and bereavement (Martin and Turnbull 2001, Foley, Timonen et al. 2012, O'Brien, Whitehead et al. 2012, Whitehead, O'Brien et al. 2012); however, these services are not routinely provided (Aoun, Connors et al. 2012, O'Brien, Whitehead et al. 2012) despite MND best practice guidelines which recommend such support (Leigh, S Abrahams et al. 2003, Mitchell and Borasio 2007). Moreover, MND family carers appear vulnerable to prolonged grief (Hebert, Lacomis et al. 2005, Aoun, Connors et al. 2012). With this in mind, the findings here that almost three-quarters of MND family carers believe the generativity document will provide comfort during bereavement is salient. Support for dignity therapy's potential to provide comfort during bereavement can be found in the comments of MND family carers, where most reported they would "treasure," reread, and reflect upon the document in the future.

Despite reports that dignity therapy was helpful to participants and that the generativity document would be helpful during bereavement, some family carers expressed ambivalence about the possible benefits of dignity therapy to themselves at

the time of the intervention. While generally expressing positive feelings about dignity therapy, some family carers also criticized that the information presented in the document was not comprehensive nor did it present anything new, and others said a document could not replace years of memories and time together. Some family carers also pointed out the intervention was not capable of alleviating the overwhelming feelings of stress, loss and grief they were experiencing. However, comments such as these are indicative of normal preparatory or anticipatory grief responses (Kubler-Ross 1969, Worden 2009) and would likely be said about any psychotherapeutic intervention implemented at this time, rather than criticisms of dignity therapy.

Interestingly, there were also some responses in this study indicating dignity therapy was not beneficial for family carers. Some family members said dignity therapy resulted in feeling confronted by death which detracted from their ability to enjoy the present. Accepting the reality of death is a primary task encountered by people facing the loss of a loved one (Worden 2009), but family members of the terminally ill commonly vacillate between tensions of denial and acceptance over time (Lynn and Harrold 1999). Several studies have documented the emotional difficulties encountered by MND family carers as a result of the confronting nature of an MND diagnosis (O'Brien, Whitehead et al. 2011a, Hogden, Greenfield et al. 2013), and research has determined that some MND family carers cope by not looking too far into the future (Bolmsjo and Hermeren 2003) or openly discussing death (Ray, Brown et al. 2012). Therefore, it is important to consider whether family members who provide support in the dignity therapy interview (and/or those who receive the generativity documents) are ready to participate in the creation and reading of a 'eulogistic' document (Schryer, McDougall et al. 2012) like that created in dignity therapy. Chochinov cautions that dignity therapy participants may not wish to acknowledge or discuss their impending death during dignity therapy, and he advises using ambiguous language around death awareness unless led by the participant to discuss death more concretely (Chochinov 2012). This caution should also be extended to involved family members, and an assessment should be made by the therapist about their level of acceptance.

Other family members said they were hurt by things the participant said during the dignity therapy interview or by the content of the generativity document. One of the

goals in the first dignity therapy meeting is to establish a "frame" for dignity therapy that includes the names and ages of important family members, and to determine who in the family will receive the dignity therapy document (Chochinov 2012 p. 75). However, what is missing is a family assessment where family systems, areas of conflict, and longstanding problems, such as substance abuse and physical abuse, are identified (Kissane, Bloch et al. 1998, Fisher 2003, Kasl-Godley, King et al. 2014). Terminal illness can add additional stressors on families, and trigger changes in roles, relationships, and communication that can increase conflict (Kasl-Godley, King et al. 2014). Moreover, families vary in their ability to communicate effectively about distressing issues, to make collaborative decisions, and to remain attached and secure through transitions and crises (Kissane, Bloch et al. 1994, Fisher 2003, Breen and O'Connor 2011). Dignity therapists require a basic knowledge of family systems, and they should assess and plan for problem areas of potential conflict, dysfunction and disagreement in order to avoid creating lasting generativity documents that can cause or add to distress in families.

In this study, almost a quarter of family carers listed negative effects at two weeks post intervention. In research by Hall et al. (2013b), two-thirds of family members reported negative effects. Hall et al. did not state at what time point family members were interviewed although it is implied it was at least four weeks post-intervention. It is possible that the increased numbers were a result of the longer period families had to consider the document. Support for this conjecture is found in Houmann et al.'s study (2014), which found as time went on, people who received dignity therapy were less confident that the document brought increased appreciation from their families. In this study, a participant and his daughters were highly satisfied with dignity therapy; however, two weeks post-intervention the participant requested changes to the document to mitigate the hurt feelings of the participant's second wife, who felt her role in his life was underplayed in the document. It is interesting that Chochinov mentions only a few instances of family member dissatisfaction with dignity therapy in his research, which he relates to cognitive impairment in the participant (Chochinov, Kristjanson et al. 2011, Chochinov 2012) and to a family member reading the document to which he or she was not supposed to be given access (Chochinov 2012). This area

warrants further inquiry. While dignity therapy was designed for people with terminal illness rather than for family carers, its impact on family members must be taken into consideration as they are part of the unit of care for palliative care providers (National Consensus Project for Quality Palliative Care 2014, World Health Organization 2014). It is important that the therapist considers the potential benefits of dignity therapy, along with potential negative effects, and proceed only if the former outweighs the latter.

## 8.4 Towards addressing 'total pain': Dignity therapy

### 8.4.1 Key findings

Best practice for evidence-based evaluation of palliative care interventions includes assessment of cost-effectiveness along with efficacy (Evans, Harding et al. 2013). In this research, dignity therapy appears to be a relatively time consuming and expensive intervention. Dignity therapy with people with MND averaged 4 sessions with client contact time of 6 hours over 42 days. The average audio recorded interview was 1 hour 38 minutes. It is estimated that the average time spent editing the interview transcripts outside the presence of the participant was about 9 hours, and the average transcription cost was $165 AUD. Previous research has documented similar findings on the average number of sessions and client contact time (Montross, Winters et al. 2011, Hall, Goddard et al. 2012), and in the total interview time, editing time and clinician time (Hall, Goddard et al. 2012). However, in other reported areas, including completion time and transcription cost, dignity therapy took longer and was more expensive to perform with people with MND than was reported in research with cancer patients (Montross, Winters et al. 2011, Johns 2013) and people in aged care (Hall, Goddard et al. 2012). It should be noted that these figures exclude dignity therapy performed by email with two participants because there was there was no oral interview, no transcription, and editing time was substantially reduced. This method was less time intensive and also did not involve time and costs associated with travel, and though the numbers are small, acceptability and effectiveness were not compromised with these participants.

Dignity therapy involved considerable time and costs that were not tracked in this or other research (Montross, Winters et al. 2011, Hall, Goddard et al. 2012)

including; travel costs and costs for supplies; and travel time, time spent arranging visits, time spent waiting, and cancelled appointments. In a palliative care environment, where many are cared for in their homes and where physical status is often unpredictable, this amount of time can be substantial. In addition, travel time and costs in WA, where the population is dispersed over a large area, were considerable. Delivery of dignity therapy to people in rural and remote areas was prohibitively time consuming, expensive and difficult to coordinate. While previous research has documented issues such as extensive editing time and frequent delays which may decrease feasibility (Passik, Kirsh et al. 2004, Hall, Goddard et al. 2012), incomplete information in previous research has made assessment of cost-effectiveness difficult.

In their dignity therapy pilot study and IRCT, Chochinov and colleagues did not provide data about the time and resources required for dignity therapy (Chochinov, Hack et al. 2005, Chochinov, Kristjanson et al. 2011). However, Chochinov does provide guidelines in instructional materials for therapists. In the *Handbook of Psychotherapy in Cancer Care* (Chochinov and McKeen 2011), Chochinov asserts that dignity therapy comprises a 1 hour interview, 3 to 4 hours to edit the transcription, and another 1 hour meeting to edit and finalize the document. This totals 2 sessions with 2 hours of patient contact time and 5 to 6 hours in total to carry out the intervention. In the book *Dignity Therapy: Final Words for Final Days* (Chochinov 2012), Chochinov provides guidelines different to his previous advice and writes that dignity therapy requires 4 or more sessions between the therapist and client: an initial meeting of 20 minutes, a 1 hour interview, a 20 minute meeting to edit the document, and a fourth, brief meeting where the final document is handed over. This totals a minimum of 4 visits with less than 2 hours of patient contact time. While providing these guidelines, Chochinov asserts "the most important factor determining the number and duration of sessions will be the patient's general state of health, energy and cognitive capacity" (Chochinov 2012 p. 95).

The reality of required clinician time appears to differ substantially from the guidelines provided by Chochinov above (Chochinov and McKeen 2011, Chochinov 2012). This study and two others (Montross, Winters et al. 2011, Hall, Goddard et al. 2012) have reported consistent findings that dignity therapy averages 4 contact sessions per participant with approximately 6 hours of contact time; however, when editing time

was included, both this study and Hall (2012) found the intervention took approximately 15 hours of therapist time per participant. Thus, the clinician hours are about 3 times greater than the guidelines presented by Chochinov in instructional materials. Moreover, Chochinov estimates the total cost of dignity therapy to be "$400 to $500" CAD per participant, which he calculates as $200 CAD spent on transcription, and 2 to 3 hours of therapist time billed at $100 CAD per hour (Chochinov 2012 p. 177). A more accurate estimate of the total cost of dignity therapy using Chochinov's transcription cost and billable rate based on research findings appears to be 3 to 4 times greater, or approximately $1,700 CAD per participant.

The mean total days to complete the intervention in this study was 42 days, which is longer than the estimate of 7 days provided by Chochinov in his manual, who advises "to aim to have all patient contacts completed in about one week" (Chochinov 2012 p. 97). An extended completion time was also found by Hall (2012), who reported a mean duration of 32 days. One can surmise that sessions and duration of the therapy would be closer to Chochinov's estimation of 7 days in people very close to death who have limited time and energy to complete the intervention; however, in the IRCT the mean survival time was 110 days (Chochinov, Kristjanson et al. 2011). Moreover, it does not appear that Chochinov intended to limit dignity therapy to people very close to death, as the manual states that dignity therapy is for "anyone facing life-threatening or life-limiting circumstances" (Chochinov 2012 p. 56) and that it "can be a welcome opportunity for anyone who wishes for a way to enhance meaning, purpose or wellbeing in their final months, weeks or days of life" (Chochinov and McKeen 2011 p. 83). Chochinov and others have also expanded the use of dignity therapy from palliative care settings into aged care settings (Chochinov, Cann et al. 2012, Hall, Goddard et al. 2012), where proximity to death is not an important consideration. These factors demonstrate that dignity therapy need not be completed quickly in cases where death is not imminent.

It should be noted that the researchers in the dignity therapy studies that have reported time and cost data, including this study, have been trained in dignity therapy by Professor Chochinov (Montross, Winters et al. 2011, Bentley, Aoun et al. 2012, Hall, Goddard et al. 2012). This fact negates differences in the delivery of the intervention due to education and training of the therapist/researcher. The exception is the study by

Johns (2013), where a health psychologist and licensed clinical social worker performed dignity therapy with cancer patients using only the manual for reference.

An unaddressed area in the literature is *who* should provide dignity therapy. The dignity therapy manual contained in the *Handbook of Psychotherapy in Cancer Care* (Chochinov and McKeen 2011) is targeted to psycho-oncologists; however, Chochinov is vague about who should perform dignity therapy in his book (Chochinov 2012) and trainings. Regular training workshops offered by Chochinov are aimed at "clinicians working in palliative care and/or geriatric patients," "researchers," and "health care and nursing home administrators" (Dignity in Care 2014 para. 5). In previous dignity therapy research, the intervention has been carried out by psychiatrists (Chochinov, Kristjanson et al. 2011, Avery and Baez 2012, Juliao, Oliveira et al. 2012), medical students (Tait and Hodges 2013), research nurses (Chochinov, Hack et al. 2005, Chochinov, Kristjanson et al. 2011), social workers (Johns 2013), social work students (Davis Berman 2014), psychologists (Chochinov, Kristjanson et al. 2011, Montross, Winters et al. 2011, Bentley, Aoun et al. 2012, Johns 2013, Houmann, Chochinov et al. 2014), and researchers who had unspecified backgrounds (Passik, Kirsh et al. 2004, Hall, Goddard et al. 2011, Hall, Goddard et al. 2012, Vaghee, Javadi et al. 2012). In clinical settings, dignity therapy appears to be conducted by a wide range of practitioners, including pastoral care providers, aged care workers, and volunteers (Squires 2014, Zahn 2014). Based on the findings of this research that dignity therapy requires specialized knowledge, skills and education, including: education in psychology and assessment; knowledge of end of life issues, symptom management, and bereavement; and skills in person-centered counseling and family therapy, it is proposed here that the practice of dignity therapy should be limited to trained mental health providers with expertise in palliative care and who have specific knowledge about the disease symptoms and trajectory of the participants with whom they work.

Despite the emerging empirical challenges to dignity therapy (Hall, Goddard et al. 2013a, Hall, Goddard et al. 2013b), and the time, costs, and difficulties sometimes encountered while delivering the intervention, dignity therapy appears to be a favored intervention often endorsed in palliative care literature and practice (Ferrell 2005, Nekolaichuk 2011, Kumar, Morse et al. 2012). There are few other palliative care

interventions where people speak with regularity about the "honor" and "privilege" of their work (Chochinov 2012 p. 99, Schryer, McDougall et al. 2012 p. 2, Tait and Hodges 2013 p. 734), or of the meaning and purpose the therapy experience has given to the practitioner (Bentley 2012, Davis Berman 2014). Chochinov says, "Of the things I have done in palliative care over the past twenty years, none have been quite as gratifying or personally engaging as Dignity Therapy" (Chochinov 2012 p. ix). It can be difficult and emotionally draining to work in palliative care, where one is regularly confronted by pain, suffering and death (Martins Pereira, Fonseca et al. 2011, Breen, O'Connor et al. 2014). In an environment where there is often little one can 'do,' dignity therapy may present an option for palliative care providers to feel they are making a difference through an intervention they perceive as positive, meaningful and enduring for the patient, the patient's family, and, less consciously, themselves. Inquiry into the benefits experienced by dignity therapists through the intervention is warranted.

## 8.5 Recommendations for therapists using dignity therapy with people with MND and MND family carers

Dignity therapy is a viable psychosocial intervention for people with MND that can be offered whenever a person has a desire to complete the intervention. Based on the support found in Hall's research (2013b) that dignity therapy addresses generativity concerns, people with MND who express a desire to document their contribution and leave messages for future generations are good candidates for the therapy. Ideally, dignity therapy would be provided by a psychotherapist who is a member of a multi-disciplinary team where information, education, and screenings about symptoms would be provided as a part of the comprehensive care provided to people with MND and their family carers. There are three main recommendations for clinicians that may be gleaned from the present study.

First, the use of screenings to assess cognitive impairment, speech impairment, and emotional lability are suggested prior to beginning dignity therapy with people with MND. The use of an MND specific cognitive screening measure, such as the ALS-CBS (Woolley, York et al. 2010), will help to determine whether participants have the cognitive capacity to complete the intervention, as well as the ability to provide

informed consent. Even with a passing score, extra care should be taken in determining the suitability of people suspected of behavioral variant FTD, who may lack the insight and empathy required to complete a generativity document that is satisfactory to family members. A screening measure, such as the CETI-m (Ball, Beukelman et al. 2004), is recommended for people who have speech impairment to determine the participant's level of speech intelligibility. Those who score below a specified level (e.g. 70% on the CETI-m) should be encouraged to use an AAC method during the dignity therapy interview. A screen for emotional lability, such as the Center for Neurologic Study-Lability Scale (CNS-LS) (Moore, Gresham et al. 1997), is recommended to help educate participants and family members, as well as inform the therapist of this potential issue. Discussion should take place with potential participants before dignity therapy begins about common MND symptoms encountered during dignity therapy and their potential effects on the participant and family carer so people with MND and their family carers have the opportunity to consider this information, speak with their health care providers if they have questions or concerns, and make informed decisions about their care.

Second, people with speech loss, significant speech impairment, and emotional lability should be given the option to complete dignity therapy via email or other computerized method. In order to serve people in rural and remote areas, using e-health methods, including email or videoconferencing, is indicated in order to address cost-effectiveness and practical issues with delivering dignity therapy. As people with MND exercise control over healthcare to adapt to their continuing losses (Foley, Timonen et al. 2014), it is argued here that giving every participant control over the decision on how to best complete dignity therapy, including using e-health methods, is appropriate whenever possible.

Third, therapists should exercise care when working with families with complex histories, such as those with multiple marriages, or those who have conflicting or estranged relationships, and endeavor to prevent harm to family members through the permanent generativity document that will be left behind. Even where complex family systems are not evident, therapists should not assume that there is no conflict or dysfunction in the family (Breen and O'Connor 2011). Though a person with MND may accept their diagnosis and wish to complete dignity therapy, therapists should also

exercise care when engaging family members who are struggling to accept the terminal nature of MND as dignity therapy may be too confronting. Therapists should also exercise caution when dealing with participants or family members from non-Western cultures as dignity therapy may be culturally incompatible for these participants and/or the recipients of the generativity document.

## 8.6 Strengths and limitations

### 8.6.1 Empirical strengths

A strength of this research is its use of a quantitative descriptive design to explore the feasibility, acceptability and potential benefits of dignity therapy for people with MND using instruments and an approach that can be replicated in future research. Prior to this study, there was no evidence for dignity therapy for people with MND and their carers. The high completion rates and recruitment of an intervention group representative of people with MND and MND family carers in demographic and health status characteristics are additional strengths of the study and provide the basis on which an RCT may be based (see section 8.7 below).

### 8.6.2 Methodological strengths

Methodological strengths include the use of MND-specific cognitive and health status measures. The self-report feedback questionnaires used to assess acceptability for both people with MND and MND family carers in this study were nearly identical to the questionnaires used in the IRCT, pilot study and other dignity therapy research, which allowed for comparisons. The use of outcomes measuring hope, spirituality, and dignity-related distress were beneficial in determining whether dignity therapy had the potential to alleviate the types of distress it was designed to address. Post-testing took place through mail or through a visit by a second researcher to mitigate response bias. The researcher was trained in dignity therapy by Professor Harvey Max Chochinov and received advice and supervision from him during the course of the research.

### 8.6.3 Clinical strengths

The people with MND in this study all had a verified diagnosis of MND, and all family carers were spouse/partners. A flexible approach to modifications to the

intervention to accommodate disease symptoms allowed for a realistic appraisal of feasibility issues, as well as the special considerations needed to perform dignity therapy with people with MND. To optimize adherence to the dignity therapy protocol, 10% of recordings, transcripts and completed documents (*N*=3) were randomly selected and reviewed by three experienced researchers (2 trained in dignity therapy and the other a Registered Psychologist) who deemed them to be adherent to the dignity therapy manualized intervention.

### 8.6.4 Limitations

The main limitations of this study were inadequate power to discover small effects, low levels of distress at baseline, and the use of outcome measures not validated for people with MND. However, the approach used was congruent with the level of evidence needed in the application of an established therapy in a novel group (Craig, Dieppe et al. 2008). While it was anticipated that the short duration of the intervention would minimize confounding variables, dignity therapy took longer to complete which may affect the validity of the findings. The study group may not be representative of people with MND as those who selected to participate may have been less distressed, more likely to think dignity therapy would be beneficial, and some may have had symptoms such as speech loss or emotional lability which discouraged participation. Likewise, the study group may not be representative of MND family carers as people with MND with severe cognitive impairment and their family carers were excluded from the study, and MND family carers who declined to participate may have been more distressed. The feedback questionnaire developed by Chochinov and colleagues, and which was used in this study, is phrased with a positive orientation and this may generate more positive responses. Limitations also include using estimations to track editing time in this study, and the inability to track reasons for non-participation with people with MND and MND family carers. Finally, the researcher also performed the intervention. As such, she was in a dual role. This created a potential threat to objectivity and the possibility that the relationship that developed between the researcher and participants during dignity therapy distorted the findings. However, this dual role is common in other dignity therapy and intervention studies. Checks and balances were maintained, including reflective journaling and ongoing professional supervision.

## 8.7 Recommendations for future research

### 8.7.1 Efficacy research

This feasibility study has set the stage for a future RCT of dignity therapy with people with MND. In addition, research with people with MND with elevated distress is warranted to explore whether low baseline levels of distress provide too little room for change on the outcome measures used or, in the alternative, whether dignity therapy is unsuitable for distressed participants. Use of an outcome measure validated to detect distress in people with MND is warranted, such as the HADS or the ALS Depression Inventory (ADI) (Ferentinos, Paparrigopoulos et al. 2011). It is also recommended that future research continues to explore the relationship between hopefulness and dignity therapy using the HHI, and examines the possible relationship between a person's level of spirituality and changes to hopefulness as a result of dignity therapy.

The IRCT allocated an equal amount of meetings and participant contact hours between the dignity therapy and client-centered care arms of the study; however, the research design did not take into account the number of therapist hours needed for editing the document that was completed outside the presence of the client and which is essential to dignity therapy. Thus, future research should focus on examining whether an equal amount of total therapist time spent on person-centered care results in differences in the outcomes. In addition, comparing dignity therapy to a self-guided expressive disclosure intervention would be helpful to determine if expression is one of the primary benefits of dignity therapy. Finally, additional research inquiring into the empirical basis of dignity therapy is warranted as there is scant evidence that dignity therapy is addressing the dignity related distress it was designed to reduce, evidenced by qualitative research (Hall, Goddard et al. 2013a, Hall, Goddard et al. 2013b) as well as no detectable reductions in distress using the PDI designed by Chochinov and colleagues, as was found in the study reported here and other studies (Chochinov, Kristjanson et al. 2011, Hall, Goddard et al. 2011, Hall, Goddard et al. 2012, Houmann, Chochinov et al. 2014).

### 8.7.2 Effects on MND family carers

MND family carers should be included in a future RCT to further explore the effects of dignity therapy on families in comparison to other treatment methods. An experimental study focusing on distressed MND family carers is needed to determine if dignity therapy has the ability to reduce anxiety and depression using the HADS. A qualitative study exploring more fully the mixed acceptability results provided in the feedback questionnaire including interviews with all family members who have read the generativity documents would increase knowledge about the potential benefits or negative effects on families of the terminally ill as a result of dignity therapy. Finally, longitudinal research is warranted to explore changing attitudes to the document over time, along with dignity therapy's ability to decrease distress on family members following bereavement. This could include ongoing qualitative interviews to track changing attitudes to dignity therapy and the generativity document.

### 8.7.3 Translational research: Cultural issues, e-health, implementation and sustainability

Future research exploring the suitability of dignity therapy for different cultural groups is indicated, including an examination of the dignity therapy question protocol within different cultures to determine if modifications to the question protocol are needed in certain instances. Given the difficulties of delivering interventions to people residing in rural areas (Breen and O'Connor 2013) and the growing impetus for cost-effective interventions at the end of life (Breen, Aoun et al. 2014), larger studies aimed at exploring the efficacy and cost-effectiveness of dignity therapy delivered through e-health is warranted, as well as future studies that focus on tracking the total time and resources required to provide dignity therapy so that the costs and benefits of dignity therapy can be evaluated. Qualitative research with clinicians who deliver dignity therapy would shed light on the challenges and perceived benefits to palliative care providers who provide dignity therapy, which could add new insight into potential costs and/or benefits of the intervention. More studies examining the themes found in generativity documents are needed, as well as comparisons of the themes discussed in documents between people of different disease groups, different genders, different ages,

and different cultures. Even though the dignity therapy questions are the same for everyone, participants emphasize different aspects of their lives in dignity therapy making the content of documents varied and unique. Discourse analysis would shed light on what people find important and meaningful, as well as what they wish to document at the end of life.

### 8.8 Closing words

People with MND and their families frequently express the need for psychological and emotional support in the literature, but the support they receive continues to focus on practical aspects of care such as nutrition, mobility, and respiratory support. Research has shown that MND family carers also need increased bereavement support, and despite being a best practice guideline, adequate bereavement support is lacking. Many people with MND and their families chose to participate in this study of dignity therapy because there were few other options available to them, and all appeared to benefit in some way simply by having someone to talk to where they could safely express themselves, who showed them unconditional positive regard and listened attentively, and where the topic of conversation did not always focus on their declining abilities and continuing losses.

This research is among the first to investigate a psychosocial intervention for use with people with MND. It answers a frequent call in the literature to test psychological and psychosocial interventions to alleviate distress and improve QOL, and is a promising step in providing an option for people with MND who desire to address areas of psychosocial and existential distress, and for palliative care providers to better address the 'total pain' of their patients. This research may lead to meaningful advancements in improving QOL and reducing distress in people with MND and their family carers, but it also indicates that dignity therapy is not suitable for, nor will it appeal to, everyone with MND. More options and opportunities for psychological, psychosocial and emotional support need to be developed. We need to do much better at providing support to people with MND and their families.

> ‘Mitch,’ he continued, softly now, ‘you don’t understand. I *want* to tell you about my life. I want to tell you before I can’t tell you anymore.’
>
> His voice dropped to a whisper. ‘I *want* someone to hear my story. Will you?’ (Albom 1997 p. 63).

Words spoken by Morrie Schwartz, a retired professor and a man with MND, to Mitch Albom, his former student and journalist, in Albom’s memoir “Tuesdays with Morrie.”

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# Appendix A

**Statements of Contribution of Co-Authors**

Statement of Contribution

I, Dr. Moira O'Connor, was involved with the project entitled *Dignity therapy: A psychotherapeutic intervention to enhance the end of life experience for people with motor neurone disease and their family carers.*

I am the primary supervisor on the project. I was involved with the research design and project coordination. I supervised the research and participated in writing the papers published as:

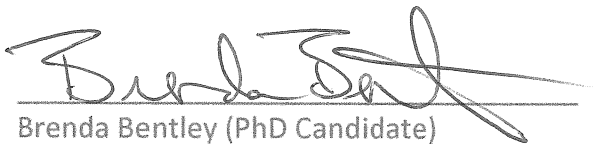
**Bentley B, Aoun SM, O'Connor M, Breen LJ, Chochinov HM (2012) Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers? *BMC Palliative Care*; 11(1):18 doi: 10.1186/1472-684X-11-18**

**Bentley B, O'Connor M, Kane R, Breen LJ (2014) Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease. *PLoS ONE* 9(5); e96888.doi: 10.1371/journal.pone.0096888;**

**Bentley B, O'Connor M, Breen LJ, Kane R (2014) Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neurone disease *BMC Palliative Care*; 13:12 doi: 10.1186/1472-684X-13-12**

The writing of this paper was led by Brenda Bentley. I commented upon drafts of this paper and checked the final version of the article.

Dr. Moira O'Connor (Co-Author)



Brenda Bentley (PhD Candidate)

Statement of Contribution

I, Dr. Lauren J. Breen, was involved with the project entitled *Dignity therapy: A psychotherapeutic intervention to enhance the end of life experience for people with motor neurone disease and their family carers.*

I co-supervised the research and participated in writing the papers published as:

**Bentley B, Aoun SM, O'Connor M, Breen LJ, Chochinov HM (2012) Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers? *BMC Palliative Care*; 11(1):18 doi: 10.1186/1472-684X-11-18**

**Bentley B, O'Connor M, Kane R, Breen LJ (2014) Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease. *PLoS ONE* 9(5); e96888.doi: 10.1371/journal.pone.0096888;**

**Bentley B, O'Connor M, Breen LJ, Kane R (2014) Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neurone disease *BMC Palliative Care*; 13:12 doi: 10.1186/1472-684X-13-12**

The writing of these papers was led by Brenda Bentley. I commented upon drafts of this paper and checked the final version of the article.

Lauren Breen

Dr. Lauren J. Breen (Co-Author)

Brenda Bentley (PhD Candidate)

Statement of Contribution

I, Dr. Robert Kane, was involved with the project entitled *Dignity therapy: A psychotherapeutic intervention to enhance the end of life experience for people with motor neurone disease and their family carers.*

I assisted with the data analysis and interpretation. I participated in writing the papers published as:

**Bentley B, O'Connor M, Kane R, Breen LJ (2014) Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease. *PLoS ONE* 9(5); e96888.doi: 10.1371/journal.pone.0096888;**

**Bentley B, O'Connor M, Breen LJ, Kane R (2014) Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neurone disease *BMC Palliative Care*; 13:12 doi: 10.1186/1472-684X-13-12**

The writing of this paper was led by Brenda Bentley. I commented upon drafts of this paper and checked the final version of the article.

Dr. Robert Kane (Co-Author)

Brenda Bentley (PhD Candidate)

Statement of Contribution

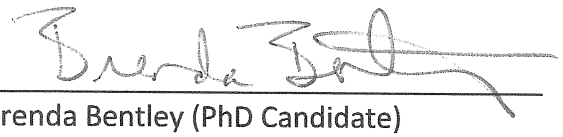
I, Professor Samar Aoun, was involved with the project entitled *Dignity therapy: A psychotherapeutic intervention to enhance the end of life experience for people with motor neurone disease and their family carers.*

I secured the scholarship funding for the study, liaised with the industry partner and led the study conception. I participated in the research design, assisted with the conduct of the study and supervised the research for three years as the primary supervisor. I participated in writing the paper published as:

**Bentley B, Aoun SM, O'Connor M, Breen LJ, Chochinov HM (2012) Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers? *BMC Palliative Care*; 11(1):18 doi: 10.1186/1472-684X-11-18**

The writing of this paper was led by Brenda Bentley. I commented upon drafts of this paper and checked the final version of the article.

Professor Samar Aoun (Co-Author)



Brenda Bentley (PhD Candidate)

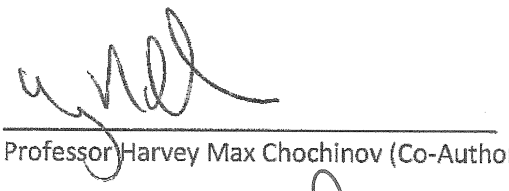
Statement of Contribution

I, Professor Harvey Max Chochinov, was involved with the project entitled *Dignity therapy: A psychotherapeutic intervention to enhance the end of life experience for people with motor neurone disease and their family carers.*

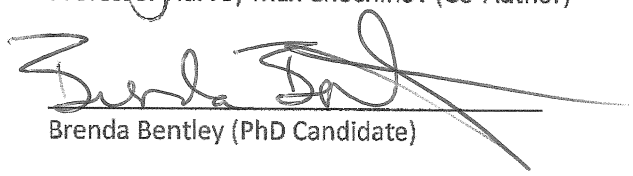
I assisted with securing the funding for the study and was involved in the study conception and design. I was initially involved in supervising the research. I participated in writing the paper published as:

**Bentley B, Aoun SM, O'Connor M, Breen LJ, Chochinov HM (2012) Is dignity therapy feasible to enhance the end of life experience for people with motor neurone disease and their family carers? *BMC Palliative Care*; 11(1):18 doi: 10.1186/1472-684X-11-18**

The writing of this paper was led by Brenda Bentley. I commented upon drafts of this paper and checked the final version of the article.



Professor Harvey Max Chochinov (Co-Author)



Brenda Bentley (PhD Candidate)

# Appendix B

**Table B-1: Recent developments in dignity therapy research**

## Table B-1: Recent developments in dignity therapy research

| Study Area | Population | Method | Country | Findings | Citation |
|---|---|---|---|---|---|
| **Feasibility** | Elderly in Aged Care | Quantitative | UK | A phase II RCT with 60 older people in care homes found no significant differences on the outcome measures, but the dignity therapy group reported more benefits on a feedback questionnaire. Dignity therapy took longer than usual to perform in this setting. | Hall, S., C. Goddard, D. Opio, P. Speck and I. J. Higginson (2012). "Feasibility, acceptability and potential effectiveness of dignity therapy for older people in care homes: A phase II randomized controlled trial of a brief palliative care psychotherapy." Palliative Medicine 26(5): 703-712 |
| **Feasibility** | Elderly in Aged Care (including dementia) | Quantitative | Canada | A feasibility study with 23 elderly people in Canada contained both cognitively intact people who completed the therapy themselves, and people with dementia who completed the therapy through a family member proxy. Participants, family and health care providers reported benefits from dignity therapy in a feedback questionnaire. | Chochinov, H. M., B. Cann, K. Cullihall, L. Kristjanson, M. Harlos, S. E. McClement, T. F. Hack and T. Hassard (2012). "Dignity therapy: A feasibility study of elders in long-term care." Palliative & Supportive Care 10(1): 3-15 |
| **Feasibility** | Terminally ill who receive the therapy via telemedicine | Quantitative | USA | Eight terminally ill people received dignity therapy using videoconferencing. Telemedicine was found to be feasible, especially with those in rural and remote locations. Participants reported high satisfaction through a feedback survey. | Passik, S. D., K. I. Kirsh and S. Leibee (2004). "A feasibility study of dignity psychotherapy delivered via telemedicine." Palliative & Supportive Care 2: 149-155 |
| **Effectiveness** | Advanced Cancer | Quantitative | UK | A phase II RCT with 45 advanced cancer patients found difference in only one outcome - hope levels were increased in the intervention group using the Herth Hope Index. The dignity therapy group also reported more benefits than the control group in a feedback survey. | Hall, S., C. Goddard, D. Opio, P. Speck, P. Martin and I. J. Higginson (2011). "A novel approach to enhancing hope in patients with advanced cancer; a randomised phase II trial of dignity therapy." BMJ Supportive and Palliative Care 1: 315-321 |
| **Effectiveness** | Advanced Cancer | Quantitative | Denmark | A pre/post study with 80 people with advanced cancer reported benefits in a feedback survey given at T1 (when the document was received). On the other outcomes, quality of life decreased and depression increased between T1 and T2 (two weeks later), and two items (sense of dignity and sense of being a burden) improved. Patients with children and those with lower emotional | Houmann L, Chochinov HM, Kristjanson L, Peterson M, Groenvald M. A prospective evaluation of Dignity Therapy in advanced cancer patients admitted to palliative care. Palliative Medicine. 28(5): 448-458 |

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| | | | | functioning and quality of life appeared to benefit more from dignity therapy. | |
| **Effectiveness** | Advanced Cancer | Quantitative | USA | Dignity therapy was completed with four participants who reported benefits in a feedback survey; however, the study had a completion rate of only 40% which is significantly lower than other studies. Those who were more distressed were more likely to drop out, indicating dignity therapy may not be ideal for distressed people. | Johns, S. (2013). "Translating dignity therapy into practice: effects and lessons learned." Omega 67(1-2): 135-145. |
| **Effectiveness** | Elderly in Aged Care | Qualitative | UK | Some people who had dignity therapy perceived some benefits for themselves and their families. The benefits did not relate to the theoretical model of dignity such as generativity, role preservation, aftermath concerns, etc. Residents from both groups perceived benefits from taking part in the study due to interactions with researchers. The dignity therapy document created problems for some who felt it was not thorough enough or worried about how their document would be perceived. Cognitive impairment and memory lapses appeared to impact on the acceptability of the document. | Hall, S., C. Goddard, P. Speck and I. J. Higginson (2013). "'It makes me feel that I'm still relevant': A qualitative study of the views of nursing home residents on dignity therapy and taking part in a phase II randomised controlled trial of a palliative care psychotherapy." Palliative Medicine 27(4): 358-366 |
| **Effectiveness** | Terminally ill with high anxiety and depression | Quantitative | Portugal | An RCT with 60 terminally ill patients with high baseline levels of anxiety and depression found that dignity therapy reduces anxiety and depression at 4 and 15 days post-intervention, though these effects were not sustained at 30 days. | Juliao, M., F. Oliveira and A. Barbosa (2012). "Efficacy of Dignity Therapy in the Anxiety of Terminally III Patients: Randomized Controlled Trial." Journal of Palliative Care 28(3): 235-235. |
| **Effectiveness** | People with chronic renal failure | Quantitative | Iran | Seventy people undergoing dialysis for chronic renal failure in Iran took part in an RCT of dignity therapy. Hope levels using the Herth Hope Index increased significantly in the dignity therapy group. Findings suggest dignity therapy may be helpful to people with chronic illness who experience hopelessness. | Vaghee, S., R. Javadi, S. R. Mazlom, N. Davoudi and M. Gharavi (2012). "The Effect of Dignity Therapy on Hope Level in Patients with Chronic Renal Failure Undergoing Hemodialysis." Life Science Journal 9(4): 3722-3727 |
| **Effectiveness:**<br><br>**Family members** | Advanced Cancer | Quantitative | USA | Six family members reported a high level of benefit from dignity therapy to themselves and their ill family member in a feedback questionnaire. | Johns, S. (2013). "Translating dignity therapy into practice: effects and lessons learned." Omega 67(1-2): 135-145. |

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| **Effectiveness:**<br><br>**Family members** | Advanced Cancer | Qualitative | UK | Interviews with 9 family members were primarily positive about dignity therapy, though 6 family members also described negative experiences. | Hall, S., C. Goddard, P. Speck, P. Martin and I. Higginson (2013). ""It makes you feel that somebody is out there caring": a qualitative study of intervention and control participants' perceptions of the benefits of taking part in an evaluation of dignity therapy for people with advanced cancer." Journal of Pain and Symptom Management 45(4): 712-725 |
| **Effectiveness:**<br><br>**Family members** | Elderly in Aged Care | Qualitative | UK | Interviews with 14 family members of people who received dignity therapy found that family members were very positive about the benefits of dignity therapy for both participants and family members who received documents. | Goddard, C., P. Speck, P. Martin and S. Hall (2013). "Dignity Therapy for older people in care homes: a qualitative study of the views of residents and recipients of 'generativity' documents." Journal of Advanced Nursing 69(1): 122-132 |
| **Cultural** | Danish | Qualitative | Denmark | Ten palliative care professionals and twenty palliative care patients were interviewed to determine if the dignity therapy interview was acceptable to a Danish group. Dignity therapy was acceptable with some revisions to the question protocol, including reducing the emphasis on self-praise, pride in oneself and accomplishments. | Houmann, L., S. Rydahl-Hansen, H. Chochinov, L. Kristjanson and M. Groenvold (2010). "Testing the feasibility of the Dignity Therapy interview: adaptation for the Danish culture." BMC Palliative Care 9(1): 21 |
| **Cultural** | Japanese | Quantitative | Japan | An initial study with consecutive subjects endured an 86% refusal rate where subjects found dignity therapy too confronting due to cultural attitudes about death. In a second study, only those expected to benefit from the therapy were approached and eleven people took part. Feedback reported high satisfaction, but not as high as those reported in western studies. Findings suggest dignity therapy should only be offered to Japanese people who wish to use a legacy. | Akechi, T., T. Akazawa, Y. Komori, T. Morita, H. Otani, T. Shinjo, T. Okuyama and M. Kobayashi (2012). "Dignity therapy: Preliminary cross-cultural findings regarding implementation among Japanese advanced cancer patients." Palliative Medicine 26(5): 768-769 |
| **Case study** | Distressed people with advanced cancer | Case study | UK | Three highly distressed people with cancer were followed after dignity therapy. They reported at 1 and 4 weeks that dignity therapy helped them and helped their families, but the authors stress distressed people are often struggling with issues dignity therapy is not meant to address, like physical pain or social problems and this adds to the complexity of delivery dignity therapy. | Hall, S., C. Goddard, P. Martin, D. Opio and P. Speck (2013). "Exploring the impact of dignity therapy on distressed patients with advanced cancer: three case studies." Psycho-Oncology 22(8): 1748-1752. |

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| **Case study** | Person with major depression | Case study | USA | A case study of a woman with major depressive disorder was presented where the subject reported it helped her find home and improved her mood. Author suggests there are similarities between people with depression and those facing terminal illness and dignity therapy should be researched with this population. | Avery, J. and M. Baez (2012). "Dignity therapy for major depressive disorder: a case report." Journal of Palliative Medicine 15(5): 509-509 |
| **Case study** | Person with severe mental illness | Case study | USA | A case study of a man with severe mental illness was presented. Dignity therapy restored hope and helped him communicate with family members. Family said they understood him better. Author states those with chronic mental illness have much in common with those facing the end of life and dignity therapy should be researched with this population. | Avery, J. and A. Savitz (2011). "A novel use of dignity therapy." The American Journal of Psychiatry 168(12): 1340-1340. |
| **Themes** | Hospice patients | Qualitative | USA | Twenty-three transcripts were analysed for the most commonly discussed areas. In rank order they were autobiographical information, love, lessons learned, important roles, accomplishments, character traits, unfinished business, hopes, catalysts, overcoming challenges, and guidance for others. | Montross, L., K. D. Winters and S. A. Irwin (2011). "Dignity therapy implementation in a community-based hospice setting." Journal of Palliative Medicine 14(6): 729-734. |
| **Themes** | Terminally ill | Qualitative | Canada, Australia | The most common values expressed in 50 dignity transcripts from the IRCT include family, pleasure, caring, accomplishment, true friendship and rich experience. | Hack, T., S. McClement, H. Chochinov, B. Cann, T. Hassard, L. Kristjanson and M. Harlos (2010). "Learning from dying patients during their final days: life reflections gleaned from dignity therapy." Palliative Medicine 24(7): 715-723. |
| **Themes** | Terminally ill | Qualitative | Canada | Stories from twelve dignity therapy transcripts were analysed and were found to have three narrative types: evaluation narratives, transition narratives, and legacy narratives. There was an overarching theme of overcoming adversity. Other themes included heritage, maturation, social lives, illness-related concerns, personal growth through illness, facing death, messages to family to appreciate life and advice for family on moving forward. | Tait, G., C. Schryer, A. McDougall and L. Lingard (2011). "Exploring the therapeutic power of narrative at the end of life: a qualitative analysis of narratives emerging in dignity therapy." BMJ Supportive and Palliative Care 1(3): 296-300. |
| **Themes** | Terminally ill | Discourse analysis | Canada | In twelve dignity therapy transcripts, participants used "eulogistic strategies" to create discursive order out of their life events through dignity therapy. | Schryer, C., A. McDougall, G. R. Tait and L. Lingard (2012). "Creating Discursive Order at the End of Life: The Role of Genres in |

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| | | | | | Palliative Care Settings." Written Communication **29**(2): 111. |
| **Clinical perspectives** | Hospice staff | Mixed methods | USA | Eighteen hospice staff members rated dignity therapy, who felt the therapy was worthwhile, helpful, had some ability to reduce pain and suffering, and that it would help family members. In qualitative interviews, staff members reported dignity therapy was a positive experience for participants and also helped to increase staff connection to patients through reading the documents. | Montross, L., E. Meier, K. DeCervantes-Monteith, V. Vashistha and S. A. Irwin (2013). "Hospice staff perspectives on dignity therapy." Journal of Palliative Medicine 16(9): 1118-1120. |
| **Clinical perspectives** | Undergraduate social work students | Qualitative | USA | Students report performing dignity therapy was a success and a meaningful experience. Themes of the impact on students at the dignity therapy interview include "greater appreciation of life," "connection to my own family," and "service and legacy." | Davis-Berman, J. (2014). "Creating a Memory Book: Undergraduate Student Experiences With End-of-Life Interviews." Death Studies **38**(2): 85-90. |
| **Clinical perspectives** | First year medical residents | Qualitative | Canada | Residents felt the art of soliciting the patients' life story was poorly taught in medical school and that a hidden message existed in their training that learning a patient's story is not a valued domain of the physician. | Tait, G. and B. Hodges (2013). "Residents learning from a narrative experience with dying patients: a qualitative study." Advances in health sciences education 18(4): 727-743. |
| **Implementation** | U.S. Cancer Centre | Implementation study | USA | Four dignity therapy transcripts were completed. The mean transcript was 11 pages long and 5878 words. Therapists spent 3 to 4 hours editing and the average interview totalled 66 minutes. | Johns, S. (2013). "Translating dignity therapy into practice: effects and lessons learned." Omega 67(1-2): 135-145. |
| **Implementation** | Elderly in Aged Care | Quantitative | UK | The mean therapist time required to perform dignity therapy with 27 participants was 15.04. Therapists spent 8.69 mean hours to edit each document, and it took 31.81 mean days to complete the intervention. | Hall, S., C. Goddard, D. Opio, P. Speck and I. J. Higginson (2012). "Feasibility, acceptability and potential effectiveness of dignity therapy for older people in care homes: A phase II randomized controlled trial of a brief palliative care psychotherapy." Palliative Medicine 26(5): 703-712 |
| **Implementation** | Hospice patients | Implementation study | USA | Twenty-three dignity therapy transcripts were completed in a hospice setting. The mean number of sessions was 4 and dignity therapy averaged 6.3 hours of direct clinical contact. The mean transcript was 8 pages long and 2993 words. | Montross, L., K. D. Winters and S. A. Irwin (2011). "Dignity therapy implementation in a community-based hospice setting." Journal of Palliative Medicine 14(6): 729-734. |

# Appendix C

**Ethics Approvals**

Curtin University

**Memorandum**

| | |
|---|---|
| **To** | Professor Samar Aoun, WA Centre for Cancer and Palliative Care |
| **From** | A/Prof Stephan Millett, Chair, Human Research Ethics Committee |
| **Subject** | Protocol Approval **HR 19/2011** |
| **Date** | 21 April 2011 |
| **Copy** | Brenda Bentley, WA Centre for Cancer and Palliative Care<br>Dr Moira O'Connor, WA Centre for Cancer and Palliative Care |

Office of Research and Development

**Human Research Ethics Committee**

**TELEPHONE** 9266 2784
**FACSIMILE** 9266 3793
**EMAIL** hrec@curtin.edu.au

Thank you for providing the additional information for the project titled "*Dignity Therapy: A psychotherapeutic intervention to enhance the end of life experience for persons with Motor Neurone Disease and their family carers*". The information you have provided has satisfactorily addressed the queries raised by the Committee. Your application is now **approved**.

- You have ethics clearance to undertake the research as stated in your proposal.
- The approval number for your project is **HR 19/2011.** *Please quote this number in any future correspondence.*
- Approval of this project is for a period of twelve months **21-04-2011** to **21-04-2012.** To renew this approval a completed Form B (attached) must be submitted before the expiry date **21-04-2012.**
- If you are a Higher Degree by Research student, data collection must not begin before your Application for Candidacy is approved by your Faculty Graduate Studies Committee.
- The following standard statement **must be** included in the information sheet to participants:

  *This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number HR 19/2011). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/- Office of Research and Development, Curtin University, GPO Box U1987, Perth, 6845 or by telephoning 9266 2784 or by emailing hrec@curtin.edu.au.*

Applicants should note the following:

It is the policy of the HREC to conduct random audits on a percentage of approved projects. These audits may be conducted at any time after the project starts. In cases where the HREC considers that there may be a risk of adverse events, or where participants may be especially vulnerable, the HREC may request the chief investigator to provide an outcomes report, including information on follow-up of participants.

The attached **FORM B** should be completed and returned to the Secretary, HREC, C/- Office of Research & Development:
When the project has finished, or

- If at any time during the twelve months changes/amendments occur, or
- If a serious or unexpected adverse event occurs, or
- 14 days prior to the expiry date if renewal is required.
- An application for renewal may be made with a Form B three years running, after which a new application form (Form A), providing comprehensive details, must be submitted.

Regards,

A/Professor Stephan Millett
Chair Human Research Ethics Committee

Curtin University

## Memorandum

| | |
|---|---|
| To | Professor Samar Aoun, WA Centre for Cancer and Palliative Care |
| From | Miss Linda Teasdale, Manager, Research Ethics |
| Subject | **PROTOCOL APPROVAL – EXTENSION HR19/2011** |
| Date | 10 May 2012 |
| Copy | Brenda Bentley, WA Centre for Cancer and Palliative Care<br>Dr Moira O'Connor, WA Centre for Cancer and Palliative Care<br>Graduate Studies Officer, Faculty of Health Sciences |

Office of Research and Development
**Human Research Ethics Committee**

TELEPHONE 9266 2784
FACSIMILE 9266 3793
EMAIL hrec@curtin.edu.au

Thank you for keeping us informed of the progress of your research. The Human Research Ethics Committee acknowledges receipt of your Form B progress report for the project *"Dignity Therapy: A psychotherapeutic intervention to enhance the end of life experience for persons with Motor Neurone Disease and their family carers."*

Approval for this project is extended for the year to **21/04/2013.**

Your approval number remains **HR19/2011.** Please quote this number in any further correspondence regarding this project.

Please note: An application for renewal may be made with a Form B three years running, after which a new application form (Form A), providing comprehensive details, must be submitted.

Thank you.

Linda Teasdale
Manager, Research Ethics
Office of Research and Development

Curtin University

## Memorandum

| | |
|---|---|
| **To** | Professor Samar Aoun, WA Centre for Cancer and Palliative Care |
| **From** | Dr Paul Copland, Manager Research Ethics |
| **Subject** | Protocol Amendment and Extension Approval **HR 19/2011** |
| **Date** | 19 February 2013 |
| **Copy** | Brenda Bentley, WA Centre for Cancer and Palliative Care<br>Dr Moira O'Connor, WA Centre for Cancer and Palliative Care |

Office of Research and Development
**Human Research Ethics Committee**

TELEPHONE 9266 2784
FACSIMILE 9266 3793
EMAIL hrec@curtin.edu.au

Thank you for keeping us informed of the progress of your research. The Human Research Ethics Committee acknowledges receipt of your Form B progress report and indication of modifications / changes for the project *"Dignity Therapy: A psychotherapeutic intervention to enhance the end of life experience for persons with Motor Neurone Disease and their family carers"*. Your application has been ***approved***.

The Committee notes the following amendments have been approved:

1. Addition of the the following recruitment methods to reach Australians with MND:
   a. Media release prepared and disseminated by Curtin University to media outlets.
   b. Social networking (Twitter, Facebook).
2. Expanding the method of delivery of intervention to include email and video-conferencing.
3. Change in method of obtaining pre/post test assessments, demographic information and feedback forms. E-health participants would complete the questionnaires and pre and post-test measures with phone or Skype assistance and return them through mail or email.
4. Change in method of performing cognitive assessment for the purpose of establishing informed consent: Reverting to the Short BOMC used previously and performing assessment by telephone or videoconference.

Approval for this project is extended to **21-04-2015.**

Your approval has the following conditions:

(i) Annual progress reports on the project must be submitted to the Ethics Office.

(ii) Please include statement of Curtin HREC approval in the media release document.

Your approval number remains **HR19/2011.** Please quote this number in any further correspondence regarding this project.

Yours sincerely

Dr Paul Copland
Manager Research Ethics

# Appendix D

**Recruitment letters**

Motor Neurone Disease Association of WA Inc
Centre for Neurological Support
The Niche
Suite B/11 Aberdare Road
Nedlands WA 6009

Phone: (08) 9346-7355
Fax: (08) 9346-7332
Web: www.mndawa.asn.au
Email: admin@mndawa.asn.au
ABN 49 312 430 982

9 May 2011

Dear

Re: An Invitation

You are invited to participate in a research study that is jointly being conducted by Curtin University and the Motor Neurone Disease Association of WA. This is not a medical research study, but it is a feasibility study that will assess the benefits of preparing a "life manuscript" for people diagnosed with MND and their carers.

In this feasibility study, you can review those aspects of your life that are most meaningful and important to you. The discussions with the researcher will be recorded and a document created that records those important aspects of your life. You and your family will receive this document of your life's achievements as you wish to have your story recorded.

This review of one's life and the creation of a "life manuscript" is called an *intervention*, and the whole process is called Dignity Therapy. The process of creating one's life manuscript or life story may reduce anxiety and allows the participants to talk about things that might otherwise be taken for granted or not discussed. This intervention has provided positive results for people diagnosed with other life-limiting conditions, but may not be a suitable process for everyone.

Family carers can also be included in this study in order to gain their perspective of the intervention. Family carers will be invited to be involved when a person diagnosed with MND agrees to participate.

Participation in this study is voluntary and if you decide to volunteer to participate, but later decide not to continue you may withdraw at any time. If you do become a participant, you will receive a Participant Information Form that provides a clear explanation of the project, as well as a Consent Form for you to sign acknowledging your consent to participate, prior to the first meeting with the Researcher, Brenda Bentley. If you do not offer to volunteer or withdraw from the study once it has started, then we will not approach you further about this study, and you will continue to receive the full services from the association.

As this is a research project, there are some inclusion criteria that have to be met. Anyone expressing an interest in participating in this research study will need to meet the following criteria to be selected to participate. Some of the inclusion criteria are: being over 18 years of age, the ability to communicate in English, and undertake a memory test. Then, if you are selected, you will find that the study is conducted in stages over the next several months so that everyone is given the time necessary for the interviews and preparation of the document. When it is your turn, all meetings will be arranged at times and locations that are most comfortable and

convenient for you, and the entire process should be completed in five meetings of an hour or less over two to three weeks. There is no cost to participants to be involved with this study.

Please indicate your interest in participating in this study by ticking the boxes. Please keep one copy of this letter and return the other marked copy in the return addressed envelope provided. If you have decided not to participate, please also return the letter with this information, so that we know your wishes and do not bother you again.

If you have any questions about this study, please contact either the Sue Colyer or the Researcher, Brenda Bentley. Our contact details are shown below

Yours sincerely

Sue Colyer

Dr Sue Colyer

Executive Officer

Brenda Bentley

Researcher
brenda.bentley@curtin.edu.au
Mobile: 0427 737 712

Your response to this invitation.

☐ .....I would like more information. *Please provide your phone number (home or mobile whichever is the easiest for contacting you) and Brenda will call to talk with you about the study.*

☐ I'm not sure. Please contact me again in six months.

☐ .....No, I do not wish to participate.

☐ Yes, I wish to participate *If you decide to participate, please indicate which is the best month for you-* May ☐ June ☐ July ☐

Return date:

Please make sure that you include both pages of this letter when you return your response to us, on or before 1 June 2011Thank you.

WA CENTRE for CANCER and PALLIATIVE CARE

Curtin University
CURTIN HEALTH INNOVATION RESEARCH INSTITUTE

Motor Neurone Disease Association of WA Inc
Centre for Neurological Support
The Niche
Suite B/11 Aberdare Road
Nedlands WA 6009

Phone: (08) 9346-7355
Fax: (08) 9346-7332
Web: www.mndawa.asn.au
Email: admin@mndawa.asn.au
ABN 49 312 430 982

15 June 2012

Dear

Re: An Update on the Dignity Therapy/MND Study

A year ago, we sent out an invitation to participate in a research study that is jointly being conducted by Curtin University and the Motor Neurone Disease Association of WA. This is not a medical research study, but a feasibility study to assess the benefits of preparing a "life manuscript" for people diagnosed with MND and their carers.

In this study you are able to review aspects of your life that are the most meaningful and important to you in a tape recorded interview. The interview is then transcribed and edited into a final document that you may give to your family or whomever you wish.

I am writing this letter today to both to give you a report on what has been accomplished in the first year and to provide you with another opportunity to participate before the study concludes in October 2013.

In the first year, twenty-one people signed up to participate. One dropped out after the initial consultation and another was not able to participate. Nineteen people with MND and twelve family carers have participated in the study so far.

I'd like to tell you a little about this first group of diverse people who have tried Dignity Therapy. There were 13 men and 6 women. They ranged in age from 32 to 80. Four live in rural areas – as far south as Northcliffe and as far north as Geraldton. Three people were still working full-time and four had young children at home. Four people were unable to speak and participated either by using a Lightwriter, using a notepad and/or whiteboard, or by email. Several others were speech impaired. Many people personalized their documents with poetry, records of achievement, timelines of important events, and photographs.

The results have been overwhelmingly positive, both from the participants and from family. Comments that have come in have shown that the therapy has proved beneficial in a number of unique ways, from helping to document one's life story, to being a reminder of important achievements, to the interview providing an opportunity to reminisce about happy events, and much more.

In this study, both people with MND and their family carers are invited to participate in order to gain both people's thoughts about the therapy. Participation in this study is voluntary and if you decide to sign up, but later decide not to continue you may withdraw at any time.

As this is a research project, there are some inclusion criteria that have to be met. Some of the inclusion criteria are: being over 18 years of age, the ability to communicate in English, and passing a very basic memory test. Then, if you continue, you will find that the study is conducted in stages over the next several months so that everyone is given the time necessary for the interviews and preparation of the document. All meetings will be arranged at times and locations that are most comfortable and convenient for you, and the entire process should be completed in five meetings of an hour or less over two to three weeks, depending mostly on you, on your availability and on your continued desire to meet. There is no cost to participate in this study.

If you have an interest in completing a life manuscript as part of the study, please tick one of the boxes below. Please keep one copy of this letter and return the other marked copy in the return addressed envelope provided. If you have decided not to participate, please also return the letter with this information, so that we know your wishes also.

If you have any questions about this study, please contact your Care Adviser, Mathew Brown, or the Researcher, Brenda Bentley.

Yours sincerely

Mathew Brown

Executive Officer

Brenda Bentley , Researcher
brenda.bentley@curtin.edu.au
Mobile: 0427 737 712

Your response to this invitation.

☐ .....I would like more information. *Please provide your email or phone number (home or mobile whichever is the easiest for contacting you) and Brenda will contact you.*

☐ I'm not sure. Please contact me again in six months. *Please provide your email or phone number (home or mobile whichever is the easiest for contacting you) and Brenda will contact you.*

☐ .....No, I do not wish to participate.

☐ Yes, I wish to participate *If you decide to participate, please indicate which is the best time* August/Sept ☐ Sept/Oct ☐ Oct/Nov ☐

Return date:

Please make sure that you include both pages of this letter when you return your response to us, on or before 10 July 2012 if possible. Thank you.

WA CENTRE for CANCER and PALLIATIVE CARE

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Motor Neurone Disease Association of WA Inc
Centre for Neurological Support
The Niche
Suite B/11 Aberdare Road
Nedlands WA 6009

Phone: (08) 9346-7355
Fax: (08) 9346-7332
Web: www.mndawa.asn.au
Email: admin@mndawa.asn.au
ABN 49 312 430 982

1 May 2013

Dear Member,

**Re: An Update on the Dignity Therapy/MND Study**

For the last two years, the Motor Neurone Disease Association of WA has been involved in a joint research study being conducted by Curtin University. This is not a medical research study, but a feasibility study to assess the benefits of preparing a "life manuscript" for people diagnosed with MND and their carers. We are writing to new members of the association who may not have heard of this study so that they may have the opportunity to participate before the study concludes in June 2013.

In this study you are able to review aspects of your life that are the most meaningful and important to you in a tape recorded interview. The interview is then transcribed and edited into a final document that you may give to your family or whomever you wish.

Twenty five people with MND and sixteen family carers have participated in the study so far. I'd like to tell you a little about this first group of diverse people who have tried Dignity Therapy. There were 18 men and 7 women. They ranged in age from 32 to 80. Five live in rural areas – as far south as Northcliffe and as far north as Geraldton. Three people were still working full-time and four had young children at home. Five people were unable to speak and participated either by using a Lightwriter, using a notepad and/or whiteboard, or by email. Several others were speech impaired. Many people personalized their documents with poetry, records of achievement, timelines of important events, and photographs.

The results have been overwhelmingly positive, both from the participants and from family. Comments that have come in have shown that the therapy has proved beneficial in a number of unique ways, from helping to document one's life story, to being a reminder of important achievements, to the interview providing an opportunity to reminisce about happy events, and much more.

In this study, both people with MND and their family carers are invited to participate in order to gain both people's thoughts about the therapy. Participation in this study is voluntary and if you decide to sign up, but later decide not to continue you may withdraw at any time.

As this is a research project, there are some inclusion criteria that have to be met. Some of the inclusion criteria are: being over 18 years of age, the ability to communicate in English, and passing a very basic memory test. All meetings will be arranged at times and locations that are most comfortable and convenient for you, and the entire process should be completed in five meetings of an hour or less over two to three weeks, depending mostly on you, on your availability and on your continued desire to meet. There is no cost to participate in this study.

If you have an interest in completing a life manuscript as part of the study, please contact **Brenda Bentley** at **0427 737 712** (mobile), or email at brenda.bentley@curtin.edu.au. Brenda will answer any questions you may have and set up a convenient appointment for the interview. No one will contact you unless you contact them first.

If you are not interested in participating, there is nothing more to do. Participation in research is voluntary. This decision will not affect in any way the support you are receiving now or may receive in the future from the Motor Neurone Disease Association.

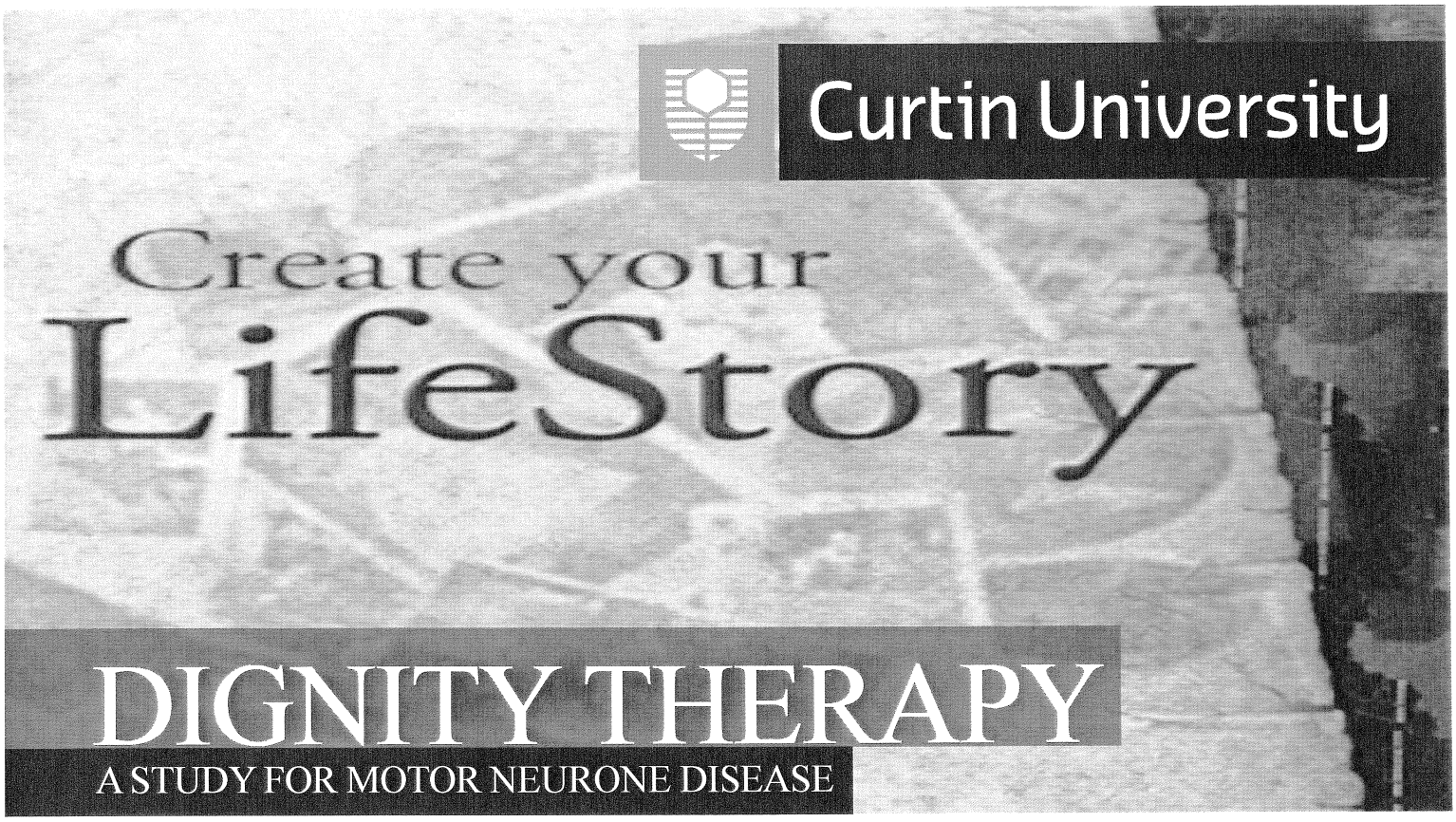
Yours sincerely

Mathew Brown
Executive Officer

Brenda Bentley, Researcher
brenda.bentley@curtin.edu.au
Mobile: 0427 737 712

# Appendix E

**Recruitment flyer**




## You are invited to take part in this study:

### Purpose of Study

The purpose of this study is to examine whether Dignity Therapy enhances the quality of life for persons with Motor Neurone Disease and their families. In previous studies, Dignity Therapy has proven to be beneficial to people with life-limiting illnesses, as well as to their family members, but most people in prior studies had cancer diagnoses. In this study, we are researching whether similar positive results will be seen with persons with Motor Neurone Disease.

### Study procedures

If you agree to take part in this study, you will receive a form of therapy called Dignity Therapy. In Dignity Therapy, you will have the opportunity to discuss things that you consider the most important or feel are or have been the most meaningful to you in life. You will be guided through a series of questions in a recorded interview in order to develop a "life manuscript," the Dignity Therapy transcript, which you will be able to keep. You will be able to have control in guiding the content of the sessions, and the audio recorder may be shut off at any time during the session. The therapy and all study procedures will take place in your care setting at times that are most convenient for you. A family carer, such as a spouse, sibling or a child, is also asked to participate in the study.

### Length of Study

The study will take about 3 weeks to complete and will comprise of about 4 visits. You will be asked to complete questionnaires at the beginning of the study and again at the end. Dignity Therapy will occur during two-three sessions in between. All sessions will take approximately 45 minutes. There will be two contacts with your family carer - once at the beginning of the study and again one week after Dignity Therapy has concluded - in order to complete questionnaires

### What to do if you are interested:

- Talk to your MNDAWA Care Adviser and ask them to refer you to the study.
- Or, contact the Researcher, Brenda Bentley, directly at 0427 737 712 or email brenda.bentley@curtin.edu.au.
- In either event, the Researcher will then explain the study to you and ask if you want to participate.
- Participation is Voluntary - your decision not to take part will not affect your care now or in the future.
- Withdrawal from the study—you are free to withdraw at any time.

**Thank you for your interest**

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Curtinnovation

Telephone 0427 737 712
or email brenda.bentley@curtin.edu.au

# Appendix F

**Participant Information Sheet and Consent Form**

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# DIGNITY THERAPY/MND STUDY

## INFORMATION SHEET – PARTICIPANTS


**INVESTIGATORS**

**Brenda Bentley, PhD Research Student, MA,**
**Prof. Samar Aoun, Principal Investigator, Director, Western Australia Centre for Cancer and Palliative Care**
**Dr. Moira O'Connor, Co-Supervisor, Western Australia Centre for Cancer and Palliative Care**
**Dr. Harvey Chochinov, Associate Supervisor, Director, Manitoba Palliative Care Research Unit**


You are being asked to participate in a research study. Please take your time to review this Information Sheet and Consent Form and discuss any questions you may have with the Research Student, Brenda Bentley, ph. (08) 9266 1765, mobile 0427 737 712, or email brenda.bentley@curtin.edu.au, or with the Principal Investigator of the study, Professor Samar Aoun ph. (08) 9266 1760. You may take your time to make your decision about participating in this research study and you may discuss it with your regular doctor, friends and family. This consent form may contain words that you do not understand. Please ask for assistance if there is any information that you do not clearly understand.

**PURPOSE OF STUDY**

The purpose of the study is to examine whether Dignity Therapy enhances the quality of life for persons and their families who are living with Motor Neurone Disease. In previous studies, Dignity Therapy has proven to be beneficial to people who have terminal illness, as well as to their family members, but most people in prior studies had cancer diagnosis. In this study, we are researching whether similar positive results will be seen with persons with Motor Neurone Disease.

A total of 50 participants diagnosed with MND from Western Australia will participate in this study. A family carer of each person with MND will also be invited to participate.

**STUDY PROCEDURES**

If you agree to take part in this research, you will receive Dignity Therapy. In Dignity Therapy, you will have the opportunity to discuss things that you consider most important or feel are/have been most meaningful to you in life. You will be guided through a series of questions about issues that are most meaningful to you, to develop a 'life manuscript'. This includes a review of your life and your most important achievements. The therapy and all study procedures will take place in your home or care setting at times that are the most convenient for you.

At the beginning of the study, you will be asked to complete a series of questionnaires. It is estimated that it will take approximately 45 minutes to do this. The Research Student,

Brenda Bentley, a therapist trained in Dignity Therapy and doctoral student at Curtin University, will assist you with this task.

The second meeting will be scheduled as soon as you are able to do so, ideally no more than 24 – 48 hours after your initial session with the Research Student. This session will be digitally recorded so that you can be provided with an edited transcript of this meeting for you to keep. The recorder may be shut off at any time during the course of the session, and you will have control on guiding the content of these sessions. The length of the session will be entirely determined by your energy level, engagement in the process, and wish to proceed. The Research Student will provide you only with as much guidance or assistance as you deem necessary. This taped session will likely last about 45 minutes.

The third meeting will consist of your receiving the typed, edited transcript of your therapy session from the previous meeting. You may read the transcript yourself, or if you prefer, the Research Student will read it aloud to you. This meeting will also provide you an opportunity to discuss your wishes regarding any editorial changes (additions, deletions or any clarifications) you wish to take place. This session is estimated to take 30 to 45 minutes.

At the fourth meeting, you will be provided with the final, edited Dignity Therapy transcript. This meeting is very brief.

A second Research Officer will then contact you one week later and provide you with the opportunity to offer feedback regarding your experience and satisfaction with the Dignity Therapy approach. We will also ask you to fill in questionnaires as you did at the beginning of the study. This session will also take about 45 minutes to complete.

**Family Carer Involvement**

It is important for this study that we also have contact with a family member or significant person involved with your care to gain information regarding the meaning/impact of this study for them. The Research Student will meet with them and ask them to complete questionnaires at the time you are entered into the study. One week after completing Dignity Therapy, the second Research Officer will meet with them and ask them to complete questionnaires. Each contact with the family carer is estimated to last 20 minutes.

## RISKS AND DISCOMFORTS/BENEFITS

While it is possible that you may benefit from your participation in this study, it is not necessarily expected that you will; however, the study should contribute to a better understanding of support needed by persons with MND such as yourself. Participants and/or family carers may experience a stirring of emotions and feelings that you may wish to discuss further with a counsellor or other support person. The Research Student will be available to discuss this with you. In the event emotional distress occurs, you will be referred by the Research Student to the Emotional Support Program available through the Motor Neurone Disease Association of WA (MNDAWA) and the Special Care Adviser appointed to oversee this program. The Emotional Support Program is an ongoing program available to you through MNDAWA and you are able to take advantage of these services at any time, regardless of your participation in the study.

## COSTS/PAYMENT FOR PARTICIPATION

There will be no cost to you for participating in this study. You will not be paid for your participation in this study.

## CONFIDENTIALITY

All information collected by us will be treated as confidential in accordance with the National Health and Medical Research Council guidelines (NHMRC). This means that all information about you including the questionnaires will be recorded with a code number and not your name, and the consent form will be kept separate from all the information collected for the study. De-identified information gained from participants will be secured in a locked filing cabinet and stored for five years. Following this time period the information will be destroyed, all written material with be shredded, computer files will be erased. An exception to this is audio recordings, which will be erased at the conclusion of the study before being disposed of to protect identity in accordance with NHMRC guidelines. If the results of the study are published, you and your family member's identity will remain confidential.

An exception to confidentiality is if you or your family member disclose a serious intent to harm yourself or someone else, and then we have an obligation to disclose only that information to your health care team.

## VOLUNTARY PARTICIPATION/WITHDRAWAL FROM THE STUDY

Your decision to take part in this study is voluntary. You may refuse to participate or you may withdraw from the study at any time, without any consequence, jeopardy or prejudice to your future medical treatment.

## QUESTIONS

You are free to ask any questions that you may have about your rights as a research participant. If any questions come up during or after the study, please contact the **Research Student, Brenda Bentley, ph. (08) 9266 1765, mobile 0427 737 712, or email brenda.bentley@curtin.edu.au, or the Principal Investigator: Professor Samar Aoun ph. (08) 9266 1760.**

*This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number HR **19/2011**). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/o Office of Research and Development, Curtin University, GPO U1987, Perth, 6845 or by telephoning 9266 2784 or by emailing hrec@curtin.edu.au.*

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## DIGNITY THERAPY/MND STUDY

## PARTICIPANT CONSENT FORM

Please do not sign this consent form unless you have a chance to ask questions and have received satisfactory answers to all of your questions.

1. I have read and understood the Information Sheet-Participants and Participant Consent Form, and I freely and voluntarily agree to take part in the research study called "Dignity Therapy/MND Study."
2. I understand that I will be given a copy of the signed and dated Information Sheet-Participants and Participant Consent Form. I have received an explanation of the purpose and duration of the study and the potential risks and benefits that I might expect. I was given sufficient time and opportunity to ask questions and to reflect back on my understanding of the study to the Research Student. My questions were answered to my satisfaction.
3. I am free to withdraw from the study at any time, for any reason, and without prejudice to my future medical treatment.
4. By signing and dating this document, I am aware that none of my legal rights are being waived.
5. I understand that all people participating in this study will be asked to fill out a series of questionnaires at the beginning and the end of the study.
6. I understand that I will be referred to the services of the Emotional Support Program through the Motor Neurone Disease Association of WA if I experience any distress as a result of my participation, which are available to me on an ongoing basis regardless of my participation.
7. I understand and give permission for the Research Student to contact my family carer in order to obtain feedback from them on the meaning/impact that this intervention has had on them.
8. I understand that the Dignity Therapy interview that I will be participating in as part of this study will be audio recorded.
9. I agree to take part in this research study and for the data obtained to be published, provided my name or other identifying information is not used.

Signature of Participant: ---------- Date: ----------

Printed name of above: ----------

I confirm that I have explained the purpose and duration of this study, as well as any potential risks and benefits, to the participant whose name and signature appear above. I confirm that I believe that the participant has understood and has knowingly given their consent to participate by his/her personally dated signature.

Signature of Researcher: ---------- Date:----------

Printed name of above: ----------

# Appendix G

**Family Carer Information Sheet and Consent Form**

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Australian Research Council

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# DIGNITY THERAPY/MND STUDY

## INFORMATION SHEET – FAMILY CARERS


**INVESTIGATORS**

**Brenda Bentley, PhD Research Student, MA,**
**Prof. Samar Aoun, Principal Investigator, Director, Western Australia Centre for Cancer and Palliative Care**
**Dr. Moira O'Connor, Co-Supervisor, Western Australia Centre for Cancer and Palliative Care**
**Dr. Harvey Chochinov, Associate Supervisor, Director, Manitoba Palliative Care Research Unit**


You are being asked to participate in a research study. Please take your time to review this Information Sheet and Consent Form and discuss any questions you may have with the Research Student, Brenda Bentley, ph. (08) 9266 1765, mobile 0427 737 712, or email brenda.bentley@curtin.edu.au, or with the Principal Investigator of the study, Professor Samar Aoun ph. (08) 9266 1760. You may take your time to make your decision about participating in this research study and you may discuss it with your regular doctor, friends and family. This consent form may contain words that you do not understand. Please ask for assistance if there is any information that you do not clearly understand.

**PURPOSE OF STUDY:**

The purpose of the study is to examine whether Dignity Therapy relieves distress and enhances the quality of life for persons and their families who are living with Motor Neurone Disease. In previous studies, Dignity Therapy has proven to be beneficial to persons with terminal illness, as well as to their family members, but most people in prior studies had cancer diagnosis. In this study, we are researching whether similar positive results will be seen with persons with Motor Neurone Disease. An important piece of this study is to understand, from the perspective of family members, how Dignity Therapy may be helpful to family carers of persons with MND.

A total of 50 participants diagnosed with MND from Western Australia will participate in this study. A family carer of each person with MND will also be invited to participate.

**STUDY PROCEDURES:**

Your family member, who has MND, will receive Dignity Therapy, described below. You will meet with the Research Student, Brenda Bentley, a psychotherapist trained in Dignity Therapy and doctoral student at Curtin University, at the beginning of the study, before Dignity Therapy begins, when you will be asked to fill out brief questionnaires. This will take approximately 20 minutes. A second Research Officer will contact you one week after Dignity Therapy has been completed to ask you to share your reaction to and feelings about this form of support. You will be asked for your reaction to the Dignity Therapy transcript.

You will also be asked to complete brief questionnaires again. This contact will also take approximately 20 minutes.

**Participant Involvement**

The person with MND will receive Dignity Therapy. In the Dignity Therapy sessions, the participant will have the opportunity to discuss things that he or she considers most important or feel are or have been most meaningful in life. The participant will be guided through a series of questions about issues that are most meaningful to develop a 'life manuscript,' the Dignity Therapy document. This includes a review of his or her life and most important achievements. The therapy and all study procedures will take place in the care setting at times that are most convenient for the participant.

At the beginning of the study, the participant will be asked to complete a series of questionnaires. At the second meeting, Dignity Therapy will take place and the session will be recorded so that an edited transcript can be provided. The audio recorder may be shut off at any time during the course of the session, and the participant will have control on guiding the content of these sessions. The length of the session will be entirely determined by the participant's energy level, engagement in the process, and wish to proceed. The Research Student will provide only as much guidance or assistance as is wanted or necessary. The third meeting will consist of receiving the typed, edited transcript of the therapy session from the previous meeting, and there will be an opportunity to discuss any editorial changes (additions, deletions or any clarifications). At the fourth, the final, edited Dignity Therapy generativity document will be returned. At the final meeting, a second Research Officer will provide the participant with the opportunity to offer feedback regarding his or her experience and satisfaction with the Dignity Therapy approach. We will also ask the participant to fill in questionnaires as they did at the beginning of the study. All participant sessions are estimated to last about 45 minutes.

## RISKS AND DISCOMFORTS/BENEFITS

While it is possible that you may benefit from your participation in this study, it is not necessarily expected that you will; however, the study should contribute to a better understanding of how Dignity Therapy is helpful to persons with Motor Neurone Disease and their family carers. Participants and/or family carers may experience a stirring of emotions and feelings that you may wish to discuss further with a counsellor or other support person. The Research Student will be available to discuss this with you. In the event emotional distress occurs, you will be referred by the Research Student to the Emotional Support Program available through the Motor Neurone Disease Association of WA (MNDAWA) and the Special Care Adviser appointed to oversee this program. The Emotional Support Program is an ongoing program available to you through MNDAWA and you are able to take advantage of these services at any time, regardless of your participation in the study.

## COSTS/PAYMENT FOR PARTICIPATION

There will be no cost to you for participating in this study. You will not be paid for your participation in this study

## CONFIDENTIALITY

All information collected by us will be treated as confidential in accordance with the National Health and Medical Research Council guidelines (NHMRC). This means that all information about you, including the questionnaires, will be recorded with a code number and not your name, and the consent form will be kept separate from all the information collected for the study. De-identified information gained from participants will be secured in a locked filing

cabinet and stored for five years. Following this time period the information will be destroyed, all written material with be shredded, computer files will be erased. An exception to this is audio recordings, which will be erased at the conclusion of the study before being disposed of to protect identity in accordance with NHMRC guidelines. If the results of the study are published, you and your family member's identity will remain confidential.

An exception to confidentiality is if you or your family member disclose a serious intent to harm yourself or someone else, and then we have an obligation to disclose only that information to your health care team.

**VOLUNTARY PARTICIPATION/WITHDRAWAL FROM THE STUDY**

Your decision to take part in this study is voluntary. You may refuse to participate or you may withdraw from the study at any time. If you withdraw, your family member may still take part in the Dignity Therapy MND study if they desire to do so, and they will not be penalized or face any prejudice in their future medical treatment.

**QUESTIONS**

You are free to ask any questions that you may have about your rights as a research participant. If any questions come up during or after the study, please contact the **Research Student, Brenda Bentley, ph. (08) 9266 1765, mobile 0427 737 712, email brenda.bentley@curtin.edu.au or Principal Investigator: Professor Samar Aoun ph. (08) 9266 1760.**

*This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number HR **19/2011**). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/o Office of Research and Development, Curtin University, GPO U1987, Perth, 6845 or by telephoning 9266 2784 or by emailing hrec@curtin.edu.au.*

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## DIGNITY THERAPY/MND STUDY

## FAMILY CARER CONSENT FORM

Please do not sign this consent form unless you have a chance to ask questions and have received satisfactory answers to all of your questions.

1. I have read and understood the Information Sheet-Family Carers and Family Carer Consent Form, and I freely and voluntarily agree to take part in the research study called "Dignity Therapy/MND Study".
2. I understand that I will be given a copy of the signed and dated Information Sheet-Family Carers and Family Carer Consent Form. I have received an explanation of the purpose and duration of the study and the potential risks and benefits that I might expect. I was given sufficient time and opportunity to ask questions and to reflect back on my understanding of the study to the Research Student. My questions were answered to my satisfaction.
3. I am free to withdraw from the study at any time, for any reason, and without prejudice to my family member who has Motor Neurone Disease.
4. By signing and dating this document, I am aware that none of my legal rights are being waived.
5. I understand that all family members participating in this study will be asked to fill out a series of questionnaires at each contact point in the study.
6. I understand that I will be referred to the services of the Emotional Support Program at the Motor Neurone Disease Association of WA if I experience any distress as a result of my participation, which are available to me on an ongoing basis regardless of my participation.
7. I agree to take part in this research study and for the data obtained to be published, provided my name or other identifying information is not used.
8. I have been assured that my name, address and telephone number will be kept confidential.
9. By signing and dating this document, I am aware that none of my legal rights are being waived.

Signature of Family Carer: ---------- Date: ----------

Printed name of above:----------

I confirm that I have explained the purpose and duration of this study, as well as any potential risks and benefits, to the participant whose name and signature appears above. I confirm that I believe that the participant has understood and has knowingly given their consent to participate by his/her personally dated signature.

Signature of Researcher: ---------- Date:----------

Printed name of above: ----------

# Appendix H

**Amyotrophic Lateral Sclerosis-Cognitive Behavioral Screen**

**(ALS-CBS)**

# ALS CBS
# ALS Cognitive Behavioral Screen

Susan C. Woolley, Ph.D.

**Patient Id:** _____ **DOB/Age:** _____ **Gender:** _____
**Onset Date:** _____ **FVC:** _____ **Education:** _____
**Onset Region:** bulbar, arm, leg, trunk, respiratory (circle one)

☐ Mark if pt responses were written, attach sheet

*HAND PAGE 2 TO CAREGIVER.*

## Attention

a. Commands: *I am going to say some commands. Please listen carefully and then do what I say. (If patient is unable to indicate with finger, movement can be substituted with eyes, arm or other means).*
1. Point/indicate (with your finger) to the ceiling and then to your left.
2. Touch your shoulder, point to the floor, and then make a fist.

# errors 0 1+
Score (circle) 1 0

b. Mental Addition/Language: *I am going to say some phrases. I want you to tell me the number of syllables in each phrase. For example, "the table" has 3 syllables. (Repetition of each phrase is allowed once).*
1. The weather is nice. (correct response: 5) answer _____
2. Tomorrow will be sunny. (correct response: 7) answer _____

# errors 0 1+
Score (circle) 1 0
(score 0 if >20 seconds on either)

c. Eye Movements: *Saccades and Antisaccades.*
# of Correct Saccades out of 8: ____/8 Score: 8/8 =**1** points, ≤7/8 = **0** points
# of Correct Antisaccades out of 8: ____/8 Score: 8/8 =**2** points, 7/8 = **1** points, ≤ 6/8 = **0 points**

/5

## Concentration

*I am going to say some numbers. After I say them, I want you to say them to me backwards, or in reverse order. For example, if I say 3-6, you would say 6-3. (If written, do not allow pt to write forward span. Discontinue after failure on two consecutive trials).*

| | | Correct | Incorrect | | | Correct | Incorrect |
|---|---|---|---|---|---|---|---|
| 2-9 | (9-2) | __ | __ | 7-8-6-4 | (4-6-8-7) | __ | __ |
| 6-4 | (4-6) | __ | __ | 5-4-1-9 | (9-1-4-5) | __ | __ |
| 3-7-2 | (2-7-3) | __ | __ | 8-2-5-9-3 | (3-9-5-2-8) | __ | __ |
| 5-8-1 | (1-8-5) | __ | __ | 5-7-6-3-9 | (9-3-6-7-5) | __ | __ |

**Maximum Span Correct:** (Enter score) /5

## Tracking/Monitoring

a. Months: *Please say the months of the year backwards, starting with December. (circle omissions/mark repetitions & intrusions)*
**Dec Nov Oct Sep Aug Jul Jun May Apr Mar Feb Jan**

# errors 0 1 2+
Score (circle) 2 1 0

b. Alphabet: *Please say/write the alphabet for me. (mark uncorrected errors, omissions or intrusions)*
**A B C D E F G H I J K L M N O P Q R S T U V W X Y Z**

# errors 0 1+
Score (circle) 1 0

c. Alternation Task: *I want you to alternate between numbers and letters, starting with 1-A, and then 2-B, 3-C, and so on. Please continue from there, alternating between number-letter, number- letter, in order, without skipping any until I tell you to stop. (Errors: Any mistake in sequencing, i.e., 7-H, or 8-9).*
**4-D 5-E 6-F 7-G 8-H 9-I 10-J 11-K 12-L 13-M**

# errors 0 1 2
Score (circle) 2 1 0

/5

## Initiation and Retrieval

*Say (write) as many words as you can starting with the letter F, as quickly as you can, in 1 minute. (Show pt Fluency Rules) You cannot say/write the names of people, places or numbers. Please do not say/write the same word with just a different ending, like truck, trucks. (S words can be substituted for F words). Errors: repetitions, rule violations.*

1. _____ 9. _____ 17. _____
2. _____ 10. _____ 18. _____
3. _____ 11. _____ 19. _____
4. _____ 12. _____ 20. _____
5. _____ 13. _____
6. _____ 14. _____
7. _____ 15. _____
8. _____ 16. _____

# correct words >12 12-8 <8 ≤4
Score (circle): 3 2 1 0*
*plus*
# errors 0 1 2+
Score (circle): 2 1 0

*if ≤4 words, total verbal fluency score = **0** regardless of # of errors

/5

**TOTAL SCORE** /20

# ALS CBS
# ALS Cognitive Behavioral Screen

Susan C. Woolley, Ph.D.

## ALS Caregiver Behavioral Questionnaire

**These questions pertain to possible changes that you have noticed since the onset of ALS symptoms. As best you can, consider changes that are unrelated to physical weakness. For example, question #1 asks about interest in activities. If the person can no longer play tennis but still seems interested in it (i.e. talks about it, watches it on television), then you would circle 3 for no change in level of interest.**

**If the person has always had the trait in question, please respond No Change, since there has been no change over time.**

**Compared to before ALS, does he/she:**

| | No Change | Small Change | Medium Change | Large Change |
|---|---|---|---|---|
| 1. Have less interest in topics/events that used to be important to them? | 3 | 2 | 1 | 0 |
| 2. Show little emotion, or seem less responsive emotionally? | 3 | 2 | 1 | 0 |
| 3. Seem more agreeable or pleasant than in the past with fewer worries? | 3 | 2 | 1 | 0 |
| 4. Fail to think things through before acting? | 3 | 2 | 1 | 0 |
| 5. Seem more withdrawn from others but not sad? | 3 | 2 | 1 | 0 |
| 6. Get confused or distracted more easily? | 3 | 2 | 1 | 0 |
| 7. Have less ability to deal with frustration or stress? | 3 | 2 | 1 | 0 |
| 8. Seem less concerned about the feelings or concerns of others than before? | 3 | 2 | 1 | 0 |
| 9. Get angry or irritable more easily than before? | 3 | 2 | 1 | 0 |
| 10. Seem more sarcastic or childlike than before? | 3 | 2 | 1 | 0 |
| 11. Eat more or have a new preference for particular foods (i.e. sweets)? | 3 | 2 | 1 | 0 |
| 12. Have more trouble changing opinions or adapting to new situations? | 3 | 2 | 1 | 0 |
| 13. Show less judgment or more problems making good decisions (i.e. regarding safety, finances, etc)? | 3 | 2 | 1 | 0 |
| 14. Have less awareness of obvious problems or changes, or deny them? | 3 | 2 | 1 | 0 |
| 15. Have new problems with language, such as saying the wrong word more often, making up new words, or declines in spelling ability? | 3 | 2 | 1 | 0 |

TOTAL SCORE: _____/45

**The following questions relate to current symptoms, not changes over time:**

| **Do you think your loved one:** | **YES** | **NO** |
|---|---|---|
| • Seems depressed on most days? | [ ] | [ ] |
| • Seems anxious on most days? | [ ] | [ ] |
| • Seems extremely fatigued on most days? | [ ] | [ ] |
| • Suffers from unexpected crying or laughing spells? | [ ] | [ ] |

**ALS CBS**
**ALS Cognitive Behavioral Screen**

Susan C. Woolley, Ph.D.

## Eye Movement Instructions

*Saccades:* *I am going to hold my fingers up. Please keep your head straight and look at me. When I wiggle a finger, I want you to look at that finger and then look back at me* (examiner should execute this eye moment themselves to demonstrate). *Look at my finger by moving your eyes only, trying to keep your head still. Each time I wiggle a finger, look at it and then back to me.* (Do 2-3 trials with the patient as practice) *We will do that a few times. Ready?* (Do 8 random trials, pause for 1-2 seconds between each trial).

*Antisaccades:* *Good, next I am going to wiggle a finger again, but this time, I want you to look AWAY from the finger that moves. For example, if I move this finger (wiggle one) then I want you to look at the other finger, not the one that moves, ok?* (Examiner should demonstrate for patient) *Let's try it* (do 2-3 trials). *Just like before, try to keep you head still and just move your eyes. After each one, look back at me. Ready?* (Do 8 random trials, pause for 1-2 seconds between each trial).

# FLUENCY RULES

## NO NAMES OF PEOPLE

## NO NAMES OF PLACES

## NO NUMBERS

## DO NOT USE SAME WORD WITH DIFFERENT ENDING

Susan C. Woolley, Ph.D.

# ALS CBS
# ALS Cognitive Behavioral Screen

## MANUAL

### Amyotrophic Lateral Sclerosis Cognitive Behavioral Screen (ALS-CBS)

### Table of Contents



**Background and General Description**

The ALS Cognitive Behavioral Screen (ALS-CBS) is a brief measure of cognition and behavior in patients with Amyotrophic Lateral Sclerosis (ALS). Up to half of patients with ALS may develop cognitive impairment during the course of the disease [1,2], and anywhere from 3-41% of ALS meet criteria for frontotemporal dementia (FTD) [1,2]. Patients with ALS who develop these types of impairments have significantly shorter survival compared to other ALS patients and are more likely to be noncompliant with interventions [3,4]. Despite this, no standardized screening measure exists to identify ALS patients who may be cognitively or behaviorally impaired, and no tools are available for the longitudinal study of impairment specific to ALS.

Susan C. Woolley, Ph.D.

# ALS CBS
## ALS Cognitive Behavioral Screen

Standard neuropsychological test batteries are time consuming, and patients with ALS are typically unable to complete them due to physical weakness, paralysis, loss of speech, or severe respiratory compromise. Moreover, most ALS centers and neurology clinics do not have neuropsychologists trained to complete detailed testing with this unique patient population. A validated, brief, and practical measure that could be administered by a variety of clinicians would fill the current gap in assessment.

The ALS CBS is composed of two sections: cognitive and behavioral. The cognitive section includes commonly used elements of standard testing batteries, consisting of 8 tasks. It can be administered by a physician or other clinical care staff and takes approximately 5 minutes to complete. The behavioral section is composed of questions sensitive to organic brain changes. It consists of a set of questions that compare changes in personality and behavior since the onset of ALS, as well as yes/no questions about mood, pseudobulbar affect, and fatigue. It is completed by a caregiver, family member, or other informant during the same time that the patient completes the cognitive portion. This questionnaire typically takes about 2 minutes.

In addition to its clinical utility, the ALS CBS may serve as a standardized and reliable research measure. It is intended for use in longitudinal assessment in order to help answer a longstanding question about whether cognitive or behavioral impairments progress in ALS patients. More specifically, the screen may provide information as to whether patients with ALS eventually develop FTD. The ALS CBS was developed to minimize demands on speech and motor capacities, so that patients can be tested during later stages of disease. Most items can be responded to using augmentative communication devices, or mouthing.

**Development**

The construction of the ALS CBS was completed in several stages. First, literature was reviewed covering topics of cognitive impairment in ALS, behavioral abnormalities in neurologic diseases, frontal lobe dysfunction, frontal lobes tests, and FTD. Existing neurocognitive screens used in neurologic diseases like dementia were reviewed. Studies evaluating the sensitivity and specificity of various neuropsychological measures in ALS patients were reviewed [1,2,5] and appropriate items were given high preference for inclusion. Consultation with a number of neurologists and neuropsychological experts in the field of ALS and motor neuron disease (MND) occurred. A meeting of several clinicians at the 2006 meeting of the Western ALS (WALS) Research Group delineated the basic structure of the screen and recommended components for inclusion.

Most cognitive items included in the ALS CBS were chosen based on high diagnostic value for frontally-mediated cognitive impairments unique to ALS. Items were also selected if they were easy for clinicians to learn, particularly for those without prior experience with neuropsychological screening tools. Items were preferentially selected if they were brief and not

highly correlated with education. Items requiring minimal motor and speech involvement were also given relative preference.

Behavioral questions were chosen based on diagnostic criteria for FTD available at the time of initial development [6] as well as initial behavioral research in the field of ALS [7,8]. Existing behavioral screens are not tailored to assess patients with ALS, who suffer from prominent and progressive physical and respiratory disability. As a result, items developed for this measure were worded to minimize the chance of endorsement based simply on disease progression. For example, apathy items were worded to assess attitudes, interest, and intellectual engagement rather than physical activity, which naturally decreases along the disease course.

Yes/No items were included on the behavioral section to track for possible mood disorders, pseudobulbar affect, and fatigue. These were intended to serve as simple screens and research data points and not as a means of diagnosis. Such items were included based on evidence that mood, emotional lability and fatigue can correlate with cognitive and behavioral changes [9-11].

An initial draft of the ALS CBS was administered to 150 consecutive ALS patients at two multidisciplinary ALS centers. All screens were reviewed by the author for accuracy and consistency. The mean cognitive score among all ALS patients was 16.3 (2.85) (total possible score: 20) versus 18.3 (1.0) in controls.

The initial draft of the ALS CBS was also administered to 24 ALS patients from the ALSA Clinic at Baylor College of Medicine with confirmed diagnoses based on cognitive and behavioral data (M=15.3; SD=4.12; 9 unimpaired, 3 cognitively impaired, 7 behaviorally impaired, and 5 FTD) [12]. A one-way analysis of variance revealed a significant difference between diagnostic classifications on the total score of the cognitive portion of the screen ($p=0.02$). The cognitive screen distinguished significantly between the cognitive classifications as scores for ALS patients diagnosed with FTD were significantly lower than both the unimpaired ($p<0.01$) and the cognitively impaired ($p=0.04$) groups; a significant difference between FTD patients and those classified as behaviorally impaired was not found ($p=0.17$). Additionally, a subset of 11 patients received both the cognitive screen (M = 15.8: SD=3.60) and a comprehensive neuropsychological battery within 9 months of each other. The neuropsychological battery assessed multiple domains including memory, visuospatial skills, language, behavior, and executive function. The validation of the cognitive section of the ALS CBS was investigated in this subset of patients with a K-means cluster analysis that resulted in a 91% correct identification of cognitively impaired patients.

York and colleagues [12] also examined the behavior portion of the screen in 24 ALS patients whose caregivers completed both the behavioral portion of the screen (M=19.2; SD=3.64) and the full Frontal Systems Behavior Scale (FrSBe) (M T-Score=62.3; SD=16.8). The behavioral

screen significantly correlated with the FrSBe Total T score (p=0.01), indicating a strong association between the behavioral screen (ALS CBS) and the standardized behavioral measure. A one-way analysis of variance revealed a significant difference between the behaviorally impaired and unimpaired diagnostic classifications with behaviorally impaired ALS patients (ALSbi) performing worse than their unimpaired counterparts (p=0.01). However, the behavioral screen did not accurately classify these patients into their respective categories (67%) based on K-means cluster analysis.

**Current Version**

Based on these results and feedback from experts in the field, revisions were made to the screen. More extensive instructions were added to the cognitive section to increase inter-rater reliability. For the behavioral questionnaire, additional items were included and the Likert scale was broadened in efforts to enhance the diagnostic accuracy. The initial version of the ALS CBS included parallel forms with similar but not identical cognitive items in order to minimize practice effects over time. However, analysis of initial data suggested that minimal practice effects occurred, even when using the same screen version over three month intervals. As a result, the second version of the ALS CBS includes only one version.

Results of the initial validation study were published in 2010 [13]. Initial analyses of data from 112 ALS patients suggest that ALS CBS™ cognitive scores were correlated with education (p=0.01), the ALS CBS behavior score (p=0.016) and forced vital capacity (FVC) (p=0.04). Behavioral scores were correlated with the cognitive score of the ALS-CBS™ and trended towards a significant correlation with FVC (p=0.06). The correlation between standardized change scores (Total Current T score- Total Premorbid T score) from the Frontal System Behavioral Scale (FrSBe) Family Rating Form and the ALS-CBS behavioral score was significant (p=0.0026).

Thirty-one ALS patients completed both the ALS-CBS and a neuropsychological assessment battery. These patients had an average age of 56 years, mean education of 14.5 years, an average FVC of 84% and average ALS FRS-R score of 34/48. The majority of patients were limb-onset (66%). Patients were separated into diagnostic classifications of cognitively and behaviourally normal (N=14), cognitively impaired (ALSci; N=8), behaviourally impaired (ALSbi; N=6) and ALS patients meeting criteria for FTD (ALS-FTD; N=6) based on results of comprehensive neuropsychological testing.

No differences existed between the ALS CBS scores of normal controls and ALS patients who were characterized as cognitively normal based on neuropsychological testing (p=0.355). A significant difference was present on cognitive scores between ALS patients with and without ALS-FTD (p=0.0005) (Table 1).

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Table 1: Means and standard deviations of ALS-CBS scores of the initial validation cohort (N=31)

| Cohort | Cognitive Score Mean (SD) Total score: 20 | Behavior Score Mean (SD) Total score: 45 |
|---|---|---|
| Normal controls (non-ALS) (N=15) | 18.8 (1.3) | 42.3 (2.1) |
| ALS cognitively & behaviourally normal (N=14,10)† | 17.7 (1.9) | 40.3 (3.8) |
| ALS cognitive impairment (ALSci) (N=8,5) | 16.1 (1.8) | 40.8 (4.4) |
| ALS behavioural impairment (ALSbi) (N=6,4) | 16.0 (3.2) | 30.2 (12.1) |
| ALS-FTD (N=6) | 3.67 (3.44)** | 22.8 (10.6)* |

†First number is sample size for cognitive test, second number is sample size for behavioural test. Some patients were included in both the ALSci and ALSbi groups depending on which diagnostic criteria they met.

**p=0.0005 (FTD vs ALS cognitively normal)

*p=0.005 (FTD vs ALS behaviourally normal)

**Comparison of screen scores across diagnostic groups**

Mean cognitive scores for the ALS normal, ALSci, ALSbi and ALSbici groups were not statistically different (p=0.35 by Kruskal-Wallis test), but the number of patients in each group was small. We did find a significant decrease in both total cognitive and total behaviour scores with advancing degrees of cognitive impairment. Linear regression of total cognitive and behavioral scores by diagnostic category (separated by controls, ALS normal, ALS impaired, and ALS-FTD) was significant (p<0.001) for each regression.

In direct group comparisons, we found differences in cognitive scores between ALS-normal (N=14) and ALS-FTD patients (N=6) (p=0.0005). To determine if non-FTD patients differed from normal controls, we collapsed the remaining four diagnostic groups into a single category (ALS, non-FTD) and found that cognitive scores differed significantly from control subjects (p=0.008, Mann-Whitney test). ALS, non-FTD also had higher cognitive scores than ALS-FTD (p=0.001, Mann-Whitney test). Finally, we grouped ALS patients with any cognitive or behavioral impairment (ALS-impaired, N=10) and found that mean cognitive scores were lower than ALS-normal, but the difference did not reach significance (p=0.15). ALS-impaired did differ significantly from the ALS-FTD group (p=0.001).

ALS-CBS behavioral scores did not differ among ALS subgroups without FTD (p=0.16 by Kruskal-Wallis test). Mean behavioral scores were again collapsed into one diagnostic group (ALS, non-FTD). Behavioral score differences for the ALS, non-FTD group and controls did not reach significance (p=0.14 by Mann-Whitney test). Behavioral scores were marginally greater for the ALS, non-FTD group than for the ALS-FTD cohort (p=0.09 by Mann-Whitney test). Behavioral scores were significantly different between ALS-normal, ALS impaired and ALS-FTD (p=0.022 by Kruskal-Wallis test). The difference between ALS-normal and ALS-FTD was significant (p=0.005).

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The behavioral section was administered to a group of ALS caregivers online via the Patients Like Me™ website. Initial results of split-half reliability from 70 caregiver responses were promising (Guttman = 0.847). Cronbach's Alpha of 0.916 suggested that none of the items need to be removed [Wicks, personal communication]. Tukey's test of additivity was not violated, suggesting that a valid total score could be obtained by summing all item scores. Test-retest reliability over a one-week period was strong (0.901).

**Interpretation of Results**

This screen is intended to identify patients in need of further assessment and should not be used as a substitute for standardized testing or formal assessment. A diagnosis of FTD or other dementia should not be given solely on the basis of this screen. In a research setting, the screen is ideally meant to help categorize patients into groups based on scores.

Screen scores should be interpreted with caution in patients with advanced age (>80) or other risk factors such as significant vascular disease, head injury, or major depression. For older patients, a MMSE or other screen should be considered to rule out Alzheimer's disease or similar dementias. Cognitive CBS scores correlate with education, and therefore results for patients with limited education need to be interpreted with caution. Given the strong correlation between cognitive scores and FVC, consideration of reversible causes of cognitive impairment need to be considered for low scores, and respiratory staff should be alerted to low screen scores which may indicate altered respiratory status. Performance of patients whose primary language is not English should be interpreted with caution.

Cognitive scores: Optimal cut points for cognition and behavioral sections were identified which best differentiated between ALS-FTD and the remainder of the cohort [13]. For the cognitive section, a cutoff of equal to or below 10 had 100% sensitivity, specificity, positive predictive value (PPV), and negative predictive value (NPV) for identifying FTD. Thus, the cutoff of 10 for cognition achieved 100% accuracy.

We further assessed the optimal cutoff that could differentiate between ALS-normal and patients with any cognitive deficit (ALSci, ALSbici, FTD). For cognition, a cutoff score greater than or equal to 17 had 86% NPV and 71% specificity to exclude cognitive impairment, while scores below 17 had 85% sensitivity and 69% PPV that there would be some cognitive impairment on a full battery. The cognitive cut-off of 17 achieved 77% accuracy for detecting any cognitive impairment.

Behavioral scores: A cutoff equal to or less than 32 gave 88% sensitivity, 80% specificity, 94% PPV and 67% NPV. Overall, the cutoff of 32 for behavior achieved 86% accuracy for correctly classifying ALS patients with FTD. A score above 36 has a specificity of 86% and NPV of 92% for predicting that no behavioral impairment would be endorsed on comprehensive evaluation, while a score equal to or less than 36 has 90% sensitivity and 82% PPV for

predicting behavioral impairment. The cut-off of 36 for behavioral score achieved 86% accuracy for detecting any behavioral impairment (either ALSbi or ALS-FTD).

The mean score for ALS-FTD patients was 22.8 (standard deviation 10.6). The most common behavioral changes endorsed by caregivers of ALS-FTD patients included decreased emotional responsiveness, withdrawal without sadness, confusion or distraction, and decreased awareness/denial of problems and changes.

For ALS patients scoring in the ranges which raise suspicion of FTD, further evaluation is required. Ideally, this would be completed by a diagnostic interview which is supplemented by standardized behavioral measures including but not limited to the Cambridge Behavioral Inventory-Revised [14], Frontal Behavior Inventory [15], or the Frontal Systems Behavioral Scale [16].

**Future Validation & Revision**

Further validation against the gold standard of neuropsychological testing is ongoing. Examination of possible correlations between ALS-CBS scores and clinical features including region of disease onset, distribution of weakness at the time of testing, distribution of upper motor neuron signs, bulbar severity, respiratory functioning, and duration of disease may enhance our understanding of whether certain phenotypes are at greater relative risk for extra-motor impairments. The correlation between cognitive screen scores and respiratory function also warrants further research. Whether ALS CBS scores correlate with neuroanatomical abnormalities is not known.

The measure has been translated in other languages and validations studies in alternate languages may provide further information about the screen's relative utility. Future versions of the screen may be amended based on additional validation studies, evolving neuropsychological research in ALS, and revised diagnostic criteria for behavioral variant FTD [17].

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(inadvertently overlooked) individuals who have a common goal of accurate assessment and effective treatment of patients with ALS.

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# ALS CBS
# ALS Cognitive Behavioral Screen

## INSTRUCTIONS

**Purpose of Screen**
This screen was developed to identify ALS patients at risk for cognitive and/or behavioral impairment. It is not meant to replace neuropsychological assessment, and should not be used to diagnose ALS-Frontotemporal Dementia (FTD), ALS-cognitive impairment (ALSci) or ALS-behavioral impairment (ALSbi). Patients may be informed that the screen assesses abilities like attention and concentration, which can be affected by many variables such as poor sleep, respiratory impairment, mood, medications, or the effects of the disease on the brain. Completing the screen can help clinicians track cognitive and behavioral functioning over time and consider alterations in treatment, as needed.

**Testing Environment**
A quiet exam room free of interruption is ideal. Testing the patient alone is preferable. However if others are in the room, politely ask them not to assist the patient with his/her responses. For all tasks, gentle and positive encouragement can be provided to the patient if they want to stop in the middle of a difficult task. However, no assistance with answering can be provided by the examiner or observer.

The behavioral questionnaire (second page of the screen) should be provided to the caregiver at the start of the examination to minimize caregiver involvement in the cognitive testing.

**Verbal versus Written Format**
This screen can be completed either orally or in writing. The clinician administering the screen should use their best judgment to determine which format to use. It is recommended that bulbar patients, who may lose their speech more rapidly, use the written format across serial assessments rather than switching from verbal to written midway through the disease course. This will allow for a more reliable assessment of change over time. For those writing responses, please provide paper or eraser-board, etc. For patients who cannot speak or write, mouthing responses or using an assistive communication device is appropriate.

**Administration Time & Frequency**
The total time for screening is approximately five minutes. More time may be required for patients who are markedly fatigued or who use a communication device. Typically, patients do not need to complete the screen more frequently than every three months. Screening at least once yearly could be considered at minimum.

## COGNITIVE SECTION

Each of the 4 sub-sections described below has a minimum score of 0 and maximum score of 5. The total score for the cognitive section ranges from 0-20, with 20 being a perfect score. To calculate the score for this section, simply sum the scores for the 4 different subsections described below.

### ATTENTION (total score: 5)

a. Commands: Recite the instructions on the form and ask the patient to wait until the entire command is read before beginning their movement. If the patient cannot point, suggest that they respond by using a limb or their eyes. Minor alterations of instructions are allowed, as this task is meant to assess attention and sequencing of 2 and 3-step tasks. Do not repeat the instructions, or if you do, score a 0 but allow the patient to complete the task by repeating the command. The only exception to this is if the patient requires repetition due to hearing impairment.

   Errors include any problem with sequencing, leaving out a step in the command, or executing the wrong action. For example, for command #1, if the patient points to their right instead of their left, that would be considered an error. For command #2, if the patient touches their shoulder, then makes a fist and points with the fist to the floor, this would be counted as an error. An exception to this may be if the patient cannot easily move their hand, and an attempt to point appears made; if this is the case, the patient would not be considered incorrect. Any errors result in a score of 0 for this subsection of the Attention subtest.

b. Mental Addition/Language: Recite the instructions on the form. If the patient does not have good command of the English language or otherwise needs clarification about what a syllable is, please explain until the patient comprehends the concept. Providing more than one example is allowed until the concept is understood. Read each phrase clearly and slowly. Repetition of each phrase once is allowed, but the 20 second time limit should start before the repetition. Record the patient's response. Any incorrect calculation or a response that is not provided within 20 seconds after the initial recitation of the sentence is considered an error. Any error results in a score of 0 for this subsection of the Attention subtest.

c. Eye Movements: Sit directly in front of the patient, about 12-24 inches away, and hold up two fingers approximately 36 inches apart, at patient's eye level. For the saccades task: Wiggle one finger. Ask the patient to look at the finger that moves without moving their head, and then look back at you. The examiner should demonstrate this for the patient to ensure that they understand the task, and then have the patient execute 2-3 practice trials before starting. Randomly move either left or right finger, wait for the patient to respond

with their eyes, and pause for 1-2 seconds between each trial. Complete a total of 8 trails. If all 8 are correct, the score is 1; any error results in a 0 score.

<u>Saccade Instructions:</u> *I am going to hold my fingers up. Please keep your head straight and look at me. When I wiggle a finger, I want you to look at that finger and then look back at me* (examiner should execute this eye moment themselves to demonstrate). *Look at my finger by moving your eyes only, trying to keep your head still. Each time I wiggle a finger, look at it and then back to me.* (Do 2-3 trials with the patient as practice) *We will do that a few times. Ready?* (Do 8 random trials, pause for 1-2 seconds between each trial).

Antisaccades: Explain to the patient that he/she will now do something different. Demonstrate the task by wiggling one finger, and instruct the patient to keep their head still and to look directly at the opposite finger without looking at the finger that moves first. Once the eye movement is complete, have the patient look back at you. Complete 2-3 practice trials before starting. Record the number correct out of 8, alternating fingers randomly. Reiteration of instructions is acceptable. 8/8 correct=score of 2, 7/8 correct=score of 1, and 2 or more errors results in a score of 0.

<u>Antisaccade Instructions</u>: *Good, next I am going to wiggle a finger again, but this time, I want you to look AWAY from the finger that moves. For example, if I move this finger (wiggle one) then I want you to look at the other finger, not the one that moves, ok?* (Examiner should demonstrate for patient) *Let's try it* (do 2-3 trials). *Just like before, try to keep you head still and just move your eyes. After each one, look back at me. Ready?* (Do 8 random trials, pause for 1-2 seconds between each trial).

Scoring errors: any incorrect gaze, even if the patient immediately self-corrects. An error is also counted if a patient attempts to anticipate your move by looking before you wiggle a finger. As an untimed test, a delay in eye movements is not considered an error unless the delay seems notable (i.e. >10 seconds).

Examiners may want to copy the instructions above and print them on the back of the cognitive section of the screen for reference, or keep a copy in examination rooms along with the rules for verbal fluency (see below). Saccade instructions are included with the ALS CBS attachment (page 3).

**CONCENTRATION** (total score: 5)

Instructions: *I am going to say some numbers. After I say them, I want you to repeat (or write) them to me backwards, or in reverse order. For example, if I say 3-6, you would say (write) 6-3. Understand?* If the patient is writing responses, ask them not to write anything until you are finished stating the span. Do not allow them to write the span forwards and then backwards.

Errors: If the patient repeats the span without reversing the numbers on the first trial, correct them, reiterate the instructions, count #1 as an error, and continue to #2. Any other errors are left uncorrected and counted as errors. If the trial is incorrect but the patient then corrects themselves spontaneously, give them credit for a correct response. Prompting or repeating of spans is not allowed. Discontinue after two consecutive errors. The score is the maximum span correct, even if patient can do a span only once. For example, if a patient completes *8-7-2* (span=3), but is incorrect with *5-8-1* (span=3), *7-8-6-4* (span=4) and *2-5-9-3* (span=4), their score is 3.

**TRACKING/MONITORING** (total score: 5)

All of these tasks are untimed.

a. Months: Ask the patient to say or write the months of the year backwards, starting with December. If the patient writes their responses, do not allow them to write the months forwards first as a reference. If possible, cover up their written responses as they are written. Typically, patients who write responses may write the first letter(s) of each month instead of writing the whole word for each. Mark any uncorrected errors. Do not tell patient when an error is made. Errors include uncorrected responses out of sequence, repetitions, intrusions, or omissions. Circle omissions and write in other errors on the form. An error-free performance results in 2 points, 1 error results in 1 point, and 2+ errors receives 0 points.

b. Alphabet: Ask the patient to say/write the alphabet, at their own pace (this is not timed). Mark any uncorrected omissions, repetitions or responses out of sequence as an error. If a patient clearly makes an error due to obvious rushing, do not count it as an error. Ask them to attempt the task again at a slower rate, but if errors are made again, count these. Any errors result in a score of 0.

c. Alternation Task: Read the instructions on the form, explaining the task. Provide the example of alternating between numbers and letters, by saying *1-A, 2-B, 3-C* slowly and clearly. Ask the patient to continue on from there until you tell them to stop. Errors are counted as any mistake in sequencing (ie. 4-D, 5-F) or in losing the task rule (i.e. 4-D, E-5, or 4-D, 5, 6, 7-E). If responses are written, again try to cover up their responses as they are provided. Stop at 13-M. An error-free performance results in 2 points, 1 error results in 1 point, and 2+ errors receives 0 points.

Susan C. Woolley, Ph.D.

**INITIATION AND RETRIEVAL** (total score: 5)

Read instructions on the form to the patient. You can provide examples to clarify the task rules. For example, *If I give you the letter T, you can say truck or trim or tiny, but you do not want to say "Thomas" or "Texas" or "twenty". Also, try not to say the same word with a different ending, like truck, trucks.* Allow the patient to reference the sheet marked Fluency Rules so they are not required to memorize the task rules.

If the patient writes their responses, provide them a blank sheet of paper, and then transcribe their responses onto the screen form. You may also staple their written responses to the form and enter the score on the sheet. They may use an augmentative communication device, although their typing speed may reduce their total score. If the examiner comprehends the intended word before it is fully typed, then this can be communicated to the patient so they have more time to generate other words.

Tell the patient to begin and then record their responses for 60 seconds. If the patient does not respond within a 15-second time period, you may provide a gentle prompt such as "Keep going". Positive encouragement can be provided without distracting the patient. Record each word stated. If you cannot understand the patient due to dysarthria, write what you can and listen for repetitions of that word.

Scoring: ≥12 correct words=3 points, 8-11 correct words=2 point, <8 correct words=1 points, and ≤4 correct words=0 points for the entire subtest, regardless of rule violations. If patient recites only three words, score entire subtest as 0, but mark the number of rule violations on the form.

Errors include repetitions, rule violations (names of people, places, numbers, or the same word with a different suffix), words starting with a different letter, or non-words. Slang and curse words are not counted as errors. Mark with an X next to any errors or circle them. If >4 correct words are generated, count and score rule violations. If no errors are made and more than 4 total words are generated, the patient gets an additional 2 points. If 1 error is made, the error score is 1. If 2 or more errors are made, the error score=0.

Total the score for correct words generated (0-3) and the error score (0-2). The maximum score is 5.

**BEHAVIORAL SECTION**

It is recommended that the behavioral questionnaire be provided to the informant (caregiver, family member) at the start of the examination to minimize the involvement of the informant in the cognitive testing. Instructions are written for the caregiver on the questionnaire.

Susan C. Woolley, Ph.D.

This should not replace a diagnostic interview for depression or other mood disorders. If depression is suspected, interpret caregiver behavioral responses with caution. If significant fatigue or respiratory distress is present, also interpret apathy questions with caution. If FTD is suspected, a comprehensive interview supported by additional standardized measures of dementia is required. These can include measures like the Cambridge Behavioral Inventory-Revised (Hodges) (www.ftdrg.org), or the Frontal Behavioral Inventory (Kertesz).

Review of results

The screen, in its current form, is designed to provide a general impression to staff and not provide a diagnosis. If a neuropsychologist or other clinician is available who has experience with screening measures, they may provide general feedback to the patient.

Initial results suggest that cognitive scores ranging from 17-20 do not support the presence of clear cognitive impairment. Scores below 16 raise suspicion of cognitive impairment, and this suspicion increases significantly as scores fall below 12. Scores below 10 raise considerable suspicion for ALS-FTD or other dementia and suggest the need for further evaluation. Please see the first section of the Manual for more detailed information about the relative accuracy of different cut off scores.

Last Updated 01/27/2014

# Appendix I

**Herth Hope Index**

**(HHI)**

ID NO: _____
FC ID NO: _____
Date: _____

# HERTH HOPE INDEX

Listed below are a number of statements. Read each statement and place an [X] in the box that describes how much you agree with that statement right now.

| | Strongly Disagree | Disagree | Agree | Strongly Agree |
|---|---|---|---|---|
| 1. I have a positive outlook toward life. | | | | |
| 2. I have short and/or long range goals. | | | | |
| 3. I feel all alone. | | | | |
| 4. I can see possibilities in the midst of difficulties. | | | | |
| 5. I have a faith that gives me comfort. | | | | |
| 6. I feel scared about my future. | | | | |
| 7. I can recall happy/joyful times. | | | | |
| 8. I have deep inner strength. | | | | |
| 9. I am able to give and receive caring/love. | | | | |
| 10. I have a sense of direction. | | | | |
| 11. I believe that each day has potential. | | | | |
| 12. I feel my life has value and worth. | | | | |


1999 items 2 & 4 reworded

## SCORING INFORMATION FOR THE HERTH HOPE SCALE (HHS)

Scoring consists of summing the ratings for the subscales and for the total scale. Subscales are based on the three factors (see Table 2 in 1991 publication). Total possible points on the total scale is 90 points. The higher the score the higher the level of hope.

Note the following items need to be reversed scored: 6, 10, 13, 17, 22, 26. Score items as follows:

Never applies to me = 0
Seldom applies to me = 1
Sometimes applies to me = 2
Often applies to me = 3

HHS has been translated into Chinese, Filipino, French, German, Japanese, Norwegian, Portuguese, Spanish, Swedish, and Tai.

Herth, K. (1991). Development and refinement of an instrument to measure hope. Scholarly Inquiry for Nursing Practice: An International Journal, 5(1), 39-51.

## SCORING INFORMATION FOR THE HERTH HOPE INDEX (HHI)

Scoring consists of summing the points for the subscale and for the total scale. Subscales are based on the three factors (see Table 2 in 1992 publication). Total possible points on the total scale is 48 points. The higher the score the higher the level of hope.

Note the following items need to be reversed scored: 3, 6. Score items as follows:

Strongly Disagree = 1
Disagree = 2
Agree = 3
Strongly Agree = 4

HHI has been translated into Arabic, Brazilian, Chinese, Dutch, French, German, Hebrew, Icelandic, Italian, Japanese, Korean, Norwegian, Portuguese, Russian, Slovenian, Spanish, Swedish, Tai, and Turkish.

Herth, K. (1992). Abbreviated instrument to measure hope: Development and psychometric evaluation. Journal of Advanced Nursing, 17, 1251-1259.

TIME: Toolkit of Instruments to Measure End-of-Life Care. http://www.chcr.brown.edu/pcoc/toolkit.htm

# Appendix J

**Patient Dignity Inventory**

ID #: _____
Date: ___ / ___ / ___
(DD / MM / YY)

## The Patient Dignity Inventory

*For each item, please indicate how much of a problem or concern these have been for you within the last few days.*

1.Not being able to carry out tasks associated with daily living (e.g. washing, getting dressed).

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

2.Not being able to attend to bodily functions independently (eg. needing assistance with toileting-related activities)

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

3.Experiencing physically distressing symptoms (such as pain, shortness of breath, nausea).

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

4.Feeling that how I look to others has changed significantly.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

5.Feeling depressed.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

6.Feeling anxious.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

7.Feeling uncertain about my health.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

8.Worrying about my future.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

9.Not being able to think clearly.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

10.Not being able to continue with my usual routines.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

11.Feeling like I am no longer who I was.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

12.Not feeling worthwhile or valued.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

Chochinov et al., 2008

ID #: _____
Date: ___ / ___ / ___
(DD / MM / YY)

13. Not being able to carry out important roles (e.g. spouse, parent).

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

14. Feeling that life no longer has meaning or purpose.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

15. Feeling that I have not made a meaningful and/or lasting contribution in my life.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

16. Feeling I have 'unfinished business' (e.g. things that I have yet to say or do; things that feel incomplete)

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

17. Concern that my spiritual life is not meaningful.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

18. Feeling that I am a burden to others.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

19. Feeling that I don't have control over my life.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

20. Feeling that care needs have reduced my privacy.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

21. Not feeling supported by my community of friends and family.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

22. Not feeling supported by my health care providers.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

23. Feeling like I am no longer able to mentally cope with challenges to my health.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

24. Not being able to accept the way things are.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

25. Not being treated with respect or understanding by others.

| 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|
| Not a Problem | A slight problem | A problem | A major problem | An overwhelming problem |

Chochinov et al., 2008

# Appendix K

**Functional Assessment of Chronic Illness Therapy – Spiritual Wellbeing Scale-12**

**(FACIT-Sp-12)**

ID NO _____
DATE _____

# FACIT-Sp-12 (Version 4)

Below is a list of statements that other people with your illness have said are important. **Please circle or mark one number per line to indicate your response as it applies to the past 7 days.**

| | | Not at all | A little bit | Some-what | Quite a bit | Very much |
|---|---|---|---|---|---|---|
| Sp1 | I feel peaceful | 0 | 1 | 2 | 3 | 4 |
| Sp2 | I have a reason for living | 0 | 1 | 2 | 3 | 4 |
| Sp3 | My life has been productive | 0 | 1 | 2 | 3 | 4 |
| Sp4 | I have trouble feeling peace of mind | 0 | 1 | 2 | 3 | 4 |
| Sp5 | I feel a sense of purpose in my life | 0 | 1 | 2 | 3 | 4 |
| Sp6 | I am able to reach down deep into myself for comfort | 0 | 1 | 2 | 3 | 4 |
| Sp7 | I feel a sense of harmony within myself | 0 | 1 | 2 | 3 | 4 |
| Sp8 | My life lacks meaning and purpose | 0 | 1 | 2 | 3 | 4 |
| Sp9 | I find comfort in my faith or spiritual beliefs | 0 | 1 | 2 | 3 | 4 |
| Sp10 | I find strength in my faith or spiritual beliefs | 0 | 1 | 2 | 3 | 4 |
| Sp11 | My illness has strengthened my faith or spiritual beliefs | 0 | 1 | 2 | 3 | 4 |
| Sp12 | I know that whatever happens with my illness, things will be okay | 0 | 1 | 2 | 3 | 4 |

Completed by:_____

**FACIT-Sp12 Scoring Guidelines** (Version 4)

Instructions:*
1. Record answers in "item response" column. If missing, mark with an X
2. Perform reversals as indicated, and sum individual items to obtain a score.
3. Multiply the sum of the item scores by the number of items in the subscale, then divide by the number of items answered. This produces the subscale score.
4. **The higher the score, the better the QOL/spiritual well-being.**

| Subscale | Item Code | Reverse item? | | Item response | Item Score |
|---|---|---|---|---|---|
| **Meaning/Peace** | Sp1 | 0 | + | _____ | =_____ |
| | Sp2 | 0 | + | _____ | =_____ |
| | Sp3 | 0 | + | _____ | =_____ |
| | Sp4 | 4 | - | _____ | =_____ |
| | Sp5 | 0 | + | _____ | =_____ |
| *Score range:* 0-32 | Sp6 | 0 | + | _____ | =_____ |
| | Sp7 | 0 | + | _____ | =_____ |
| | Sp8 | 4 | - | _____ | =_____ |

***Sum individual item scores:*** _____
***Multiply by 8:*** _____
***Divide by number of items answered:*** _____ **=Meaning/Peace subscale score**

| Subscale | Item Code | Reverse item? | | Item response | Item Score |
|---|---|---|---|---|---|
| **Faith** | Sp9 | 0 | + | _____ | =_____ |
| | Sp10 | 0 | + | _____ | =_____ |
| *Score range:* 0-16 | Sp11 | 0 | + | _____ | =_____ |
| | Sp12 | 0 | + | _____ | =_____ |

***Sum individual item scores:*** _____
***Multiply by 4:*** _____
***Divide by number of items answered:*** _____ **=Faith subscale score**

**To Derive a FACIT-Sp12 total score:**
*Score range:* 0-48

_____ (**Meaning/Peace** score) + _____ (**Faith** score) =_____ **=FACIT-Sp12Total score**

*For guidelines on handling missing data and scoring options, please refer to the Administration and Scoring Guidelines in the manual or on-line at www.facit.org.

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*PROVIDING A VOICE FOR PATIENTS WORLDWIDE*

June 16, 2014

This letter serves as permission for Brenda Bentley to include the FACIT-Sp-12 questionnaire and scoring instructions in the appendix of her PhD thesis.

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Elmhurts, IL 60126
USA
jbredle@facit.org

# Appendix L

**Participant Feedback Questionnaire**

Dignity Therapy/MND Study

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

## PARTICIPANT FEEDBACK QUESTIONNAIRE

We would appreciate your feedback and impressions of *Dignity Therapy.* Please answer each of the following questions.

1. Where is the *Dignity Therapy* document kept?

2. Have you had the opportunity to read it? How often have you either read or looked at the *Dignity Therapy* document?

3. How many copies of the *Dignity Therapy* document have been made?

4. How many people have read the *Dignity Therapy* document? Who are these people?

5. Do you plan to share the *Dignity Therapy* document with others? Who are these people?

6. Have any health care providers read the *Dignity Therapy* document? What was the result?

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

7. If you have not yet shared the *Dignity Therapy* document with health care providers, are you considering doing so in the future? Why or why not?

_____

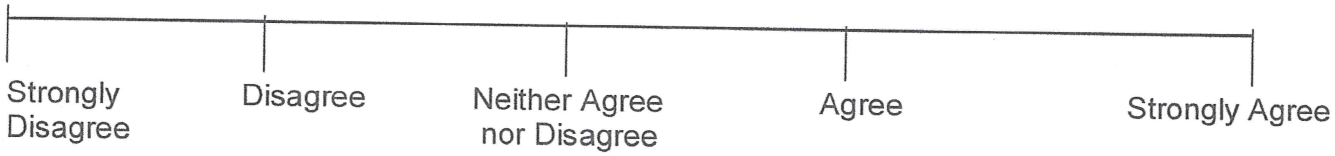
_____

_____

_____

**Please answer each of the following, choosing an answer ranging from 'strongly agree' to 'strongly disagree':**

8. I have found *Dignity Therapy* to be helpful to me.



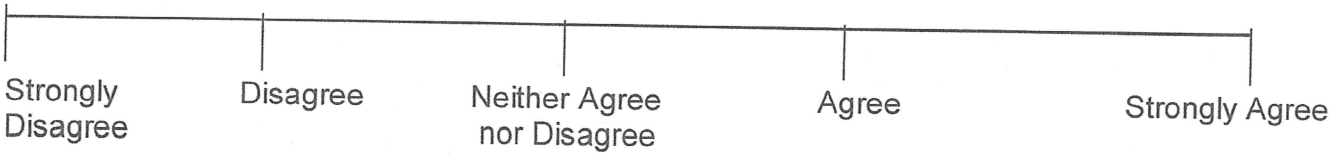

What are your reasons for that opinion?

_____

_____

_____

9. I have found *Dignity Therapy* to be as helpful as any other aspect of my health care.



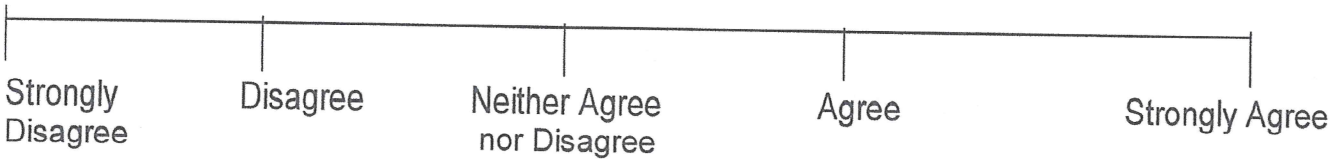

What are your reasons for that opinion?

_____

_____

_____

10. I believe *Dignity Therapy* has improved my quality of life.




What are your reasons for that opinion?

_____

_____

_____

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

11. *Dignity Therapy* has given me a sense of looking after unfinished business.

| Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____

_____

_____

12. *Dignity Therapy* has improved my sense of spiritual wellbeing.

| Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____

_____

_____

13. *Dignity Therapy* has lessened my sense of sadness or depression.

| Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____

_____

_____

14. *Dignity Therapy* has lessened my sense of feeling a burden to others.

| Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____

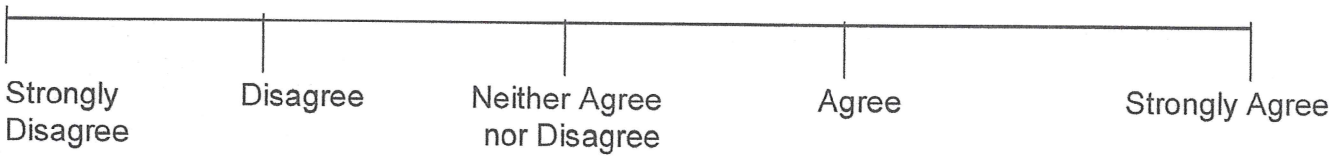
_____

_____

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

15. *Dignity Therapy* has helped me to feel more worthwhile or valued.



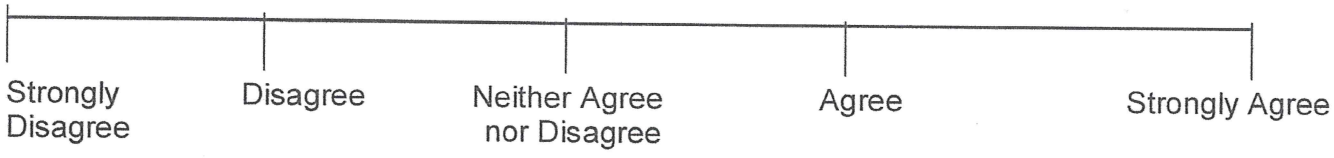

What are your reasons for that opinion?

_____

_____

_____

16. *Dignity Therapy* has helped me to feel like I am still me.



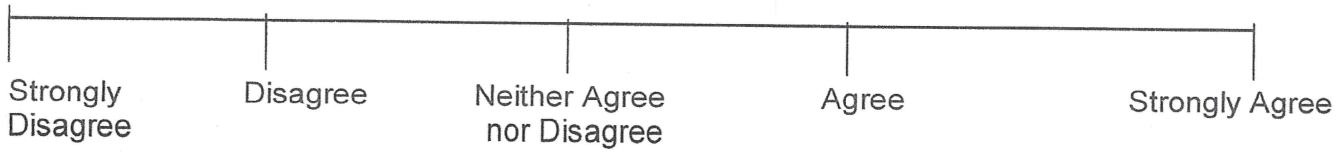

What are your reasons for that opinion?

_____

_____

_____

17. *Dignity Therapy* has given me a greater sense of having control over my life.



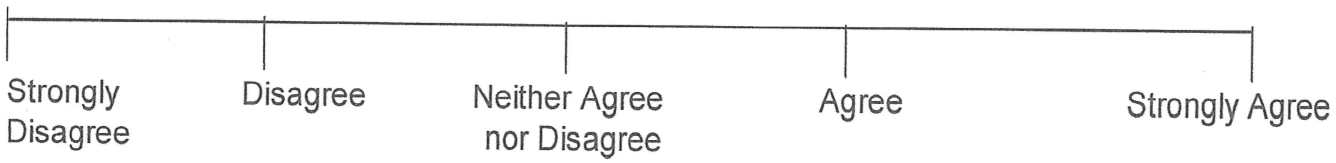

What are your reasons for that opinion?

_____

_____

_____

18. *Dignity Therapy* has helped me to accept the way things are.




What are your reasons for that opinion?

_____

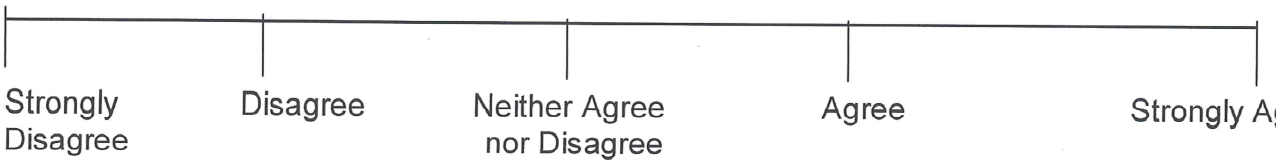
_____

_____

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

19. *Dignity Therapy* has helped me feel more respected and understood by others.



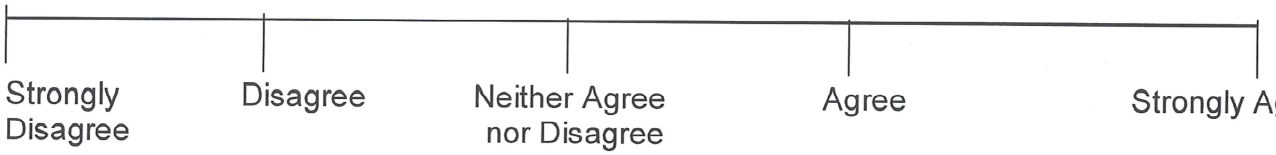

What are your reasons for that opinion?

_____

_____

_____

20. *Dignity Therapy* has helped me feel that I am still able to carry out an important task or fill an important role.



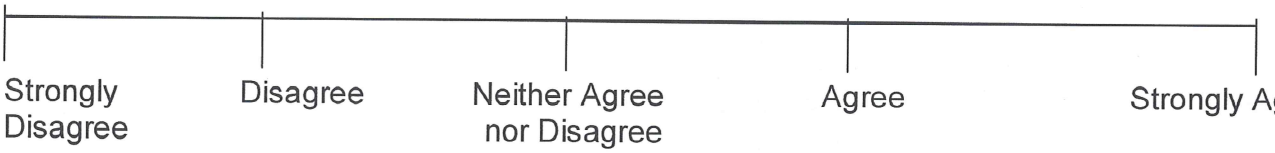

What are your reasons for that opinion?

_____

_____

_____

21. I have found *Dignity Therapy* to be satisfactory.



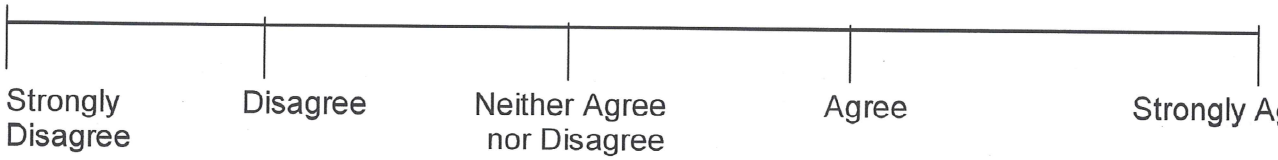

What are your reasons for that opinion?

_____

_____

_____

22. *Dignity Therapy* made me feel that my life currently is more meaningful.




What are your reasons for that opinion?

_____

_____

_____

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

23. *Dignity Therapy* has given me a heightened sense of purpose.

Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree

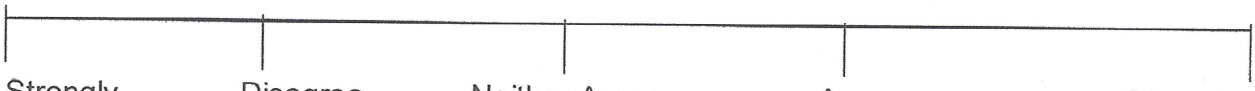
What are your reasons for that opinion?

_____

_____

_____

24. *Dignity Therapy* has given me a heightened sense of dignity.



Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree

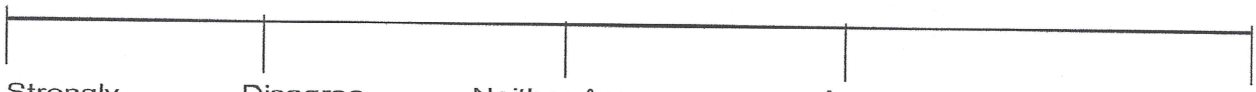
What are your reasons for that opinion?

_____

_____

_____

25. *Dignity Therapy* has lessened my sense of suffering.



Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree

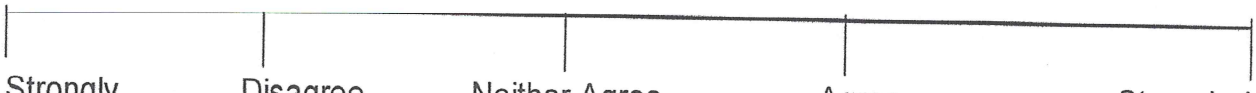
What are your reasons for that opinion?

_____

_____

_____

26. *Dignity Therapy* has made me feel more hopeful.



Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree

What are your reasons for that opinion?

_____

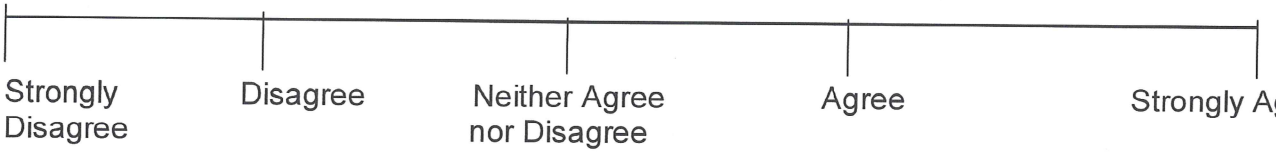
_____

_____

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

27. *Dignity Therapy* has increased my will to live.



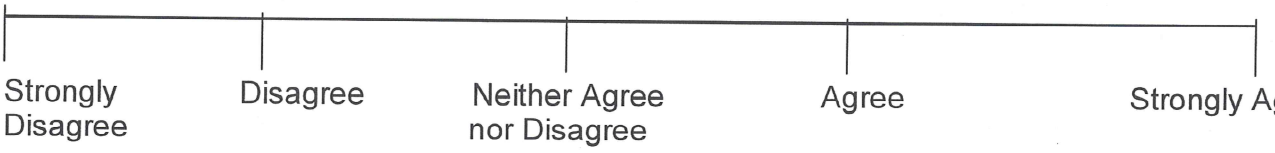

What are your reasons for that opinion?

_____

_____

_____

28. *Dignity Therapy* has helped me feel closer to the people who mean the most to me.



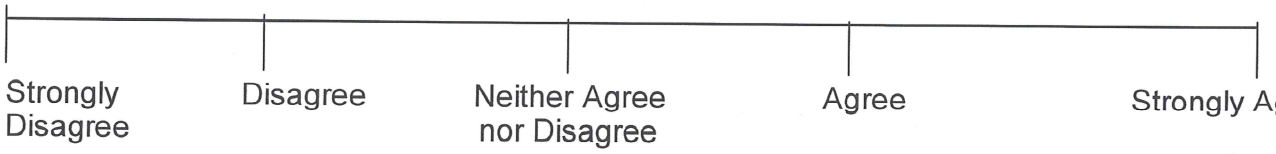

What are your reasons for that opinion?

_____

_____

_____

29. I believe *Dignity Therapy* has or will be of help to my family.




What are your reasons for that opinion?

_____

_____

_____

30. I believe *Dignity Therapy* could change the way my family sees or appreciates me.


What are your reasons for that opinion?

_____

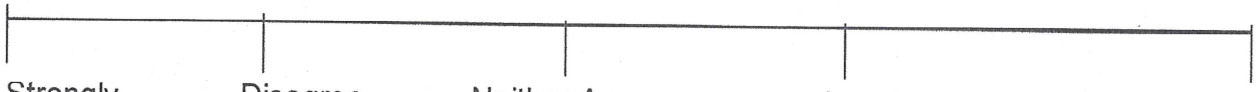
_____

_____

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

31. I believe *Dignity Therapy* could change the way my health care providers see or appreciate me.



| Strongly Disagree | Disagree | Neither Agree nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

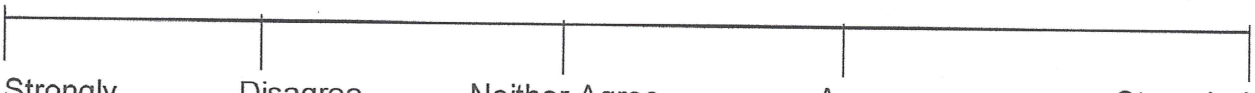
What are your reasons for that opinion?

_____

_____

_____

32. I would recommend *Dignity Therapy* to other patients and family members who are dealing with Motor Neurone Disease.



| Strongly Disagree | Disagree | Neither Agree Nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____

_____

_____

33. Do you have any other comments about how you think Dignity Therapy helped you and/or your family?

_____

_____

_____

34. Any other comments regarding your experience of *Dignity Therapy*, and how you think it might be improved, would be most appreciated.

_____

_____

_____

_____

Thank you for completing this questionnaire.

# Appendix M

**Zarit Burden Interview**

# ZARIT BURDEN INTERVIEW

INSTRUCTIONS: The following is a list of statements, which reflect how people sometimes feel when taking care of another person. After each statement, indicate how often you feel that way; never, rarely, sometimes, quite frequently or nearly always. There are no right or wrong answers.

| | Never | Rarely | Sometimes | Quite Frequently | Nearly Always |
|---|---|---|---|---|---|
| 1) Do you feel that because of the time you spend with your relative you don't have enough time for yourself? | 0 | 1 | 2 | 3 | 4 |
| 2) Do you feel stressed between caring for your relative and trying to meet other responsibilities for your family or work? | 0 | 1 | 2 | 3 | 4 |
| 3) Do you feel angry when you are around your relative? | 0 | 1 | 2 | 3 | 4 |
| 4) Do you feel that your relative's condition currently affects your relationship with other family members or friends in a negative way? | 0 | 1 | 2 | 3 | 4 |
| 5) Do you feel tense when you are around your relative? | 0 | 1 | 2 | 3 | 4 |
| 6) Do you feel your health has suffered because of your involvement with your relative? | 0 | 1 | 2 | 3 | 4 |
| 7) Do you feel that you don't have as much privacy as you would like because of your relative? | 0 | 1 | 2 | 3 | 4 |
| 8) Do you feel that your social life has suffered because you are caring for your relative? | 0 | 1 | 2 | 3 | 4 |
| 9) Do you feel you have lost control of your life since your relative's illness? | 0 | 1 | 2 | 3 | 4 |
| 10) Do you feel uncertain about what to do about your relative? | 0 | 1 | 2 | 3 | 4 |
| 11) Do you feel you should be doing more for your relative? | 0 | 1 | 2 | 3 | 4 |
| 12) Do you feel you could do a better job in caring for your relative? | 0 | 1 | 2 | 3 | 4 |

# Appendix N

**Hospital Anxiety and Depression Scale (HADS)**

**(Unable to be reproduced here due to copyright restrictions)**

# Appendix O

**Family Carer Feedback Questionnaire**

IDNO: _____
FC IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

## FAMILY CARER FEEDBACK QUESTIONNAIRE

We are interested in your feelings and reactions to *Dignity Therapy* that your family member who has MND received as a part of this study. Please read each of the following statements, fill in the lines or circle the answer that best fits with your opinion.

1. Where is the *Dignity Therapy* document kept, if you know?

_____
_____
_____

2. Have you had the opportunity to read it? How often have you either read or looked at the *Dignity Therapy* document?

_____
_____
_____

3. How many copies of the *Dignity Therapy* document have been made?

_____

4. How many people have read the *Dignity Therapy* document? Who are these people?

_____
_____
_____

5. Have any health care providers read the *Dignity Therapy* document? What was the result?

_____
_____
_____
_____

6. If your family member has not yet shared the *Dignity Therapy* document with health care providers, are they considering doing so in the future? Why or why not?

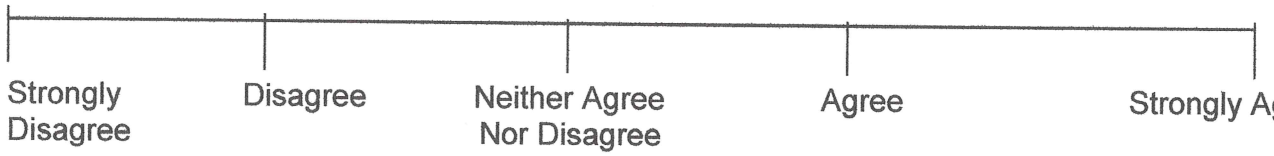
_____
_____
_____
_____

IDNO: _____
FC IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

***Your thoughts on how Dignity Therapy might have helped your family member:*** Answer each of the following, choosing an answer ranging from 'strongly agree' to 'strongly disagree'.

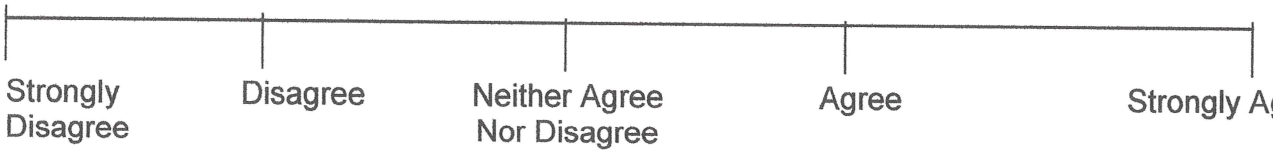
7. I believe that *Dignity Therapy* was helpful to my family member.




What are your reasons for that opinion?

_____
_____
_____

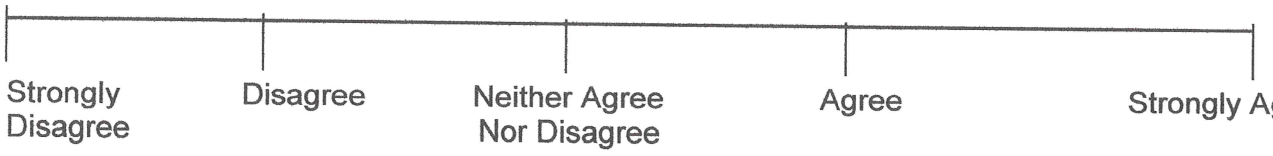
8. I believe that *Dignity Therapy* helped to give my family member a heightened sense of purpose or meaning.




What are your reasons for that opinion.

_____
_____
_____

9. I believe that *Dignity Therapy* helped to increase my family member's sense of dignity.



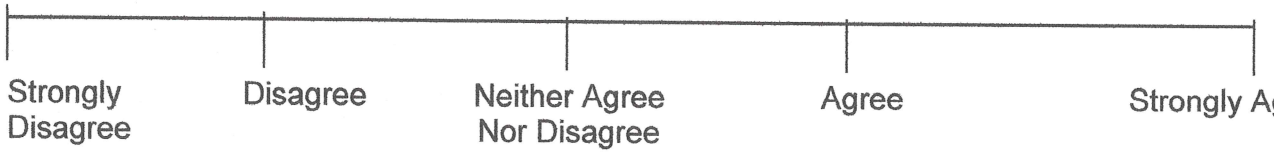

What are your reasons for that opinion?

_____
_____
_____

IDNO: _____
FC IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

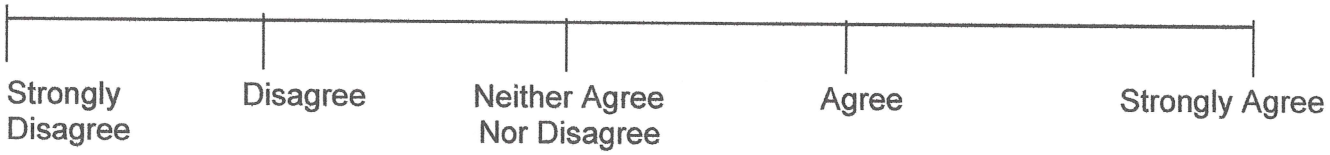
10. I believe that *Dignity Therapy* has helped prepare my family member for the end-of-life.




What are your reasons for that opinion?

_____
_____
_____

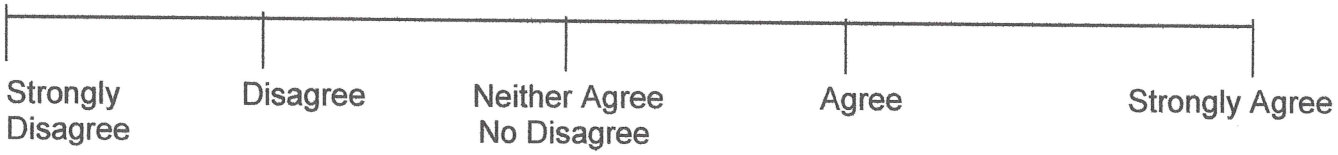
11. I believe *Dignity Therapy* was as an important a component of my family member's care as any other aspect of their care, including pain management.




What are your reasons for that opinion?

_____
_____
_____

12. I believe that *Dignity Therapy* helped to reduce my family member's suffering.




What are your reasons for that opinion?

_____
_____
_____

IDNO: _____
FC IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

13. I believe that *Dignity Therapy* helped to increase my family member's hopefulness about the future.

| Strongly Disagree | Disagree | Neither Agree No Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____
_____
_____

14. Do you have any other comments about how you think *Dignity Therapy* helped or affected your family member?

_____
_____
_____
_____
_____
_____

***Your thoughts on how Dignity Therapy might have helped you:*** Answer each of the following, choosing an answer ranging from 'strongly agree' to 'strongly disagree'.

15. The *Dignity Therapy* document helped me during this time of our life.

| Strongly Disagree | Disagree | Neither Agree Nor Agree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____
_____
_____

IDNO: _____
FC IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

16. I believe that *Dignity Therapy* has helped me to prepare for the end-of-life of my family member.

| Strongly Disagree | Disagree | Neither Agree Nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____
_____
_____

17. I believe that *Dignity Therapy* was helpful to me in reducing my feelings of stress as a family carer.

| Strongly Disagree | Disagree | Neither Agree No Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____
_____
_____

18. *Dignity Therapy* helped me feel closer to my family member.

| Strongly Disagree | Disagree | Neither Agree Nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____
_____
_____

IDNO: _____
FC IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

19. I believe that *Dignity Therapy* increased my hopefulness about the future,

| Strongly Disagree | Disagree | Neither Agree No Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____
_____
_____

20. I believe that the *Dignity Therapy* document will continue to be a source of comfort for my family and me.

| Strongly Disagree | Disagree | Neither Agree Nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____
_____
_____

21. I would recommend *Dignity Therapy* to other patients or family members who are dealing with Motor Neurone Disease.

| Strongly Disagree | Disagree | Neither Agree Nor Disagree | Agree | Strongly Agree |
|---|---|---|---|---|

What are your reasons for that opinion?

_____
_____
_____

IDNO: _____
FC IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

22. Can you think of any way we could improve *Dignity Therapy* to make it more helpful?

23. Did your family member experience any negative effects of participating in *Dignity Therapy*?

24. Did you experience any negative effects of participating in *Dignity Therapy*?

25. Do you have any other comments about how you think *Dignity Therapy* helped you and or your family?

Thank you for completing this questionnaire.

# Appendix P

**Participant Demographic and Health Questionnaire**

Dignity Therapy/MND Study

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

## PARTICIPANT DEMOGRAPHIC AND HEALTH QUESTIONNAIRE

1. What is your age? _____

2. What is your Date of Birth? (DD/MM/YY) ___/____/____

3. Gender: 1. Male 2. Female

4. Marital Status (circle one):
   0. Never Married
   1. Widowed
   2. Married/Defacto
   3. Divorced/Separated

5. Presently Living With (circle as many as apply):
   0. Alone
   1. Spouse/Partner
   2. Parent(s)
   3. Sibling(s)
   4. Child(ren)
   5. Other relative(s)
   6. Friend(s)
   7. Other (Please specify:) _____

6. Primary Social Support (circle as many as apply):
   0. Alone
   1. Spouse/Partner
   2. Parent(s)
   3. Sibling(s)
   4. Child(ren)
   5. Other relative(s)
   6. Friend(s)
   7. Other (Please specify:) _____

7. Usual Residence (circle one):
   1. Urban/Metropolitan
   2. Rural

8. What is your residence postcode? _____

9. Do you identify with a particular ethnic origin or cultural background? (circle one):
   0. No
   1. Yes If yes, please specify: _____

10. Please indicate highest level of education completed (circle one):
   0. No Formal Education
   1. Primary/Elementary School
   2. Secondary/High School
   3. University/College/Technical/Tertiary
   4. Postgraduate

Dignity Therapy/MND Study

IDNO: _____

DATE: ____/____/____ (DD/MM/YY)

11. Occupation (circle one):

1. Retired
2. Home maker
3. Unskilled (including manual workers, junior sales staff, junior clerical staff)
4. Skilled (including craftsman, technician, senior clerical, senior sales staff)
5. Manager (including middle management, small business owner, administrator)
6. Professional/Executive (including senior administrators and owners of medium to large businesses)

12. Current employment status (circle one):

0. None
1. Full-time
2. Part-time

Health History Questions:

13. Time since initial diagnosis (circle one):

1. Less than one year
2. 1 to 2 years
3. 2 to 3 years
4. 3 to 4 years
5. More than 4 years

14. Diagnosis date (if known): _____

15. Time since initial symptoms (circle one):

0. Less than one year
1. 1 to 2 years
2. 2 to 3 years
3. 3 to 4 years
4. More than 4 years

16. Date initial symptoms began (if known): _____

17. At the present time, do you have or are you being treated for any other major medical condition? (Circle as many as apply).

| | |
|---|---|
| 0. None | 5. Depression |
| 1. Heart disease | 6. Neurological condition (eg. Multiple Sclerosis, Parkinson's) |
| 2. Diabetes | 7. Arthritis |
| 3. High blood pressure | 8. Asthma |
| 4. Anxiety | 9. Other (Please specify) _____ |

18. What type of support have you had to help you cope with your illness? (Circle as many as apply).

0. None
1. Support group
2. Home care
3. Counselling
4. Respite
5. Psychologist/Psychiatrist
6. Medication
7. Other (Please specify)_____

19. If you have been prescribed medication to help you cope, please specify: (Circle as many as apply).

0. None
1. Anti-anxiety medication
2. Anti-depressant medication
3. Other (Please specify) _____

20. **In the past, prior to your MND diagnosis,** have you been diagnosed with depression? (circle one):

0. No
1. Yes

21. Do you consider yourself to be a religious person? (circle one)

0. Not at all
1. Somewhat
2. Yes, very much

22. Do you attend religious services? (circle one)

0. Never
1. Sometimes
2. Regularly

23. Do you consider yourself a spiritual person? (circle one)

0. Not at all
1. Somewhat
2. Yes, very much.

Thank you for completing this questionnaire.

# Appendix Q

**Family Carer Demographic and Health Questionnaire**

Dignity Therapy/MND Study

IDNO: _____
FC IDNO: _____
DATE: ____/____/____ (DD/MM/YY)

## FAMILY CARER DEMOGRAPHIC AND HEALTH QUESTIONNAIRE

1. What is your age? _____

2. What is your Date of Birth? (DD/MM/YY) ____/____/____

3. Gender: 1. Male 2. Female

4. Marital Status (circle one):
   0. Never Married
   1. Widowed
   2. Married/Defacto
   3. Divorced/Separated

5. Relationship to patient (circle one):
   1. Spouse/Partner
   2. Parent
   3. Sibling
   4. Adult Child
   5. Other relative(s)
   6. Friend(s)
   7. Other (please specify) _____

6. Do you reside with the patient?
   0. No
   1. Yes

7. Usual Residence (circle one):
   1. Urban/Metropolitan
   2. Rural

8. What is your residence postcode? _____

9. Do you identify with a particular ethnic origin or cultural background? (circle one):
   0. No
   1. Yes If yes, please specify: _____

10. Please indicate highest level of education completed (circle one):
   0. No Formal Education
   1. Primary/Elementary School
   2. Secondary/High School
   3. University/College/Technical/Tertiary
   4. Postgraduate

IDNO: _____
FC IDNO: _____
DATE: ____/____/____ (DD/MM/YY)

11. Occupation (circle one):
1. Retired
2. Home maker
3. Unskilled (including manual workers, junior sales staff, junior clerical staff)
4. Skilled (including craftsman, technician, senior clerical, senior sales staff)
5. Manager (including middle management, small business owner, administrator)
6. Professional/Executive (including senior administrators and owners of medium to large businesses)

12. Current employment status (circle one):
0. None
1. Full-time
2. Part-time

13. Caring hours **per day** (circle one):
1. 4 or less
2. 4 to 8
3. 8 to 12
4. More than 12

14. How long have you known the patient? (circle one)
1. Less than five years
2. Five to ten years
3. Ten to 25 years
4. More than 25 years
5. My whole life (e.g. parents, children, siblings)

Health History Questions

15. At the present time, do you have or are you being treated for any other major medical condition? (Circle as many as apply).

| | |
|---|---|
| 0. None | 5. Depression |
| 1. Heart disease | 6. Neurological condition (eg. Multiple Sclerosis, Parkinson's) |
| 2. Diabetes | 7. Arthritis |
| 3. High blood pressure | 8. Asthma |
| 4. Anxiety | 9. Other (Please specify) _____ |

Chochinov, et al, 2005 (modified)

IDNO: _____
FC IDNO: _____
DATE: ____/____/____ (DD/MM/YY)

16. What type of support have you had to help you cope with your loved one's illness? (Circle as many as apply).

0. None
1. Support group
2. Home care
3. Counselling
4. Respite
5. Psychologist/Psychiatrist
6. Medication
7. Other (please specify): _____

17. If you have been prescribed medication to help you cope, please specify: (Circle as many as apply).

0. None
1. Anti-anxiety medication
2. Anti-depressant medication
3. Other (please specify):_____

18. **In the past**, before the patient's MND diagnosis, have you been diagnosed with depression? (circle one)

0 No
1. Yes

19. Do you consider yourself to be a religious person? (circle one)

0. Not at all.
1. Somewhat.
2. Yes, very much.

20. Do you attend religious services? (circle one)

0. Never
1. Sometimes
2. Regularly

21. Do you consider yourself a spiritual person? (circle one)

0. Not at all.
1. Somewhat
2. Yes, very much.

Thank you for completing this questionnaire.

# Appendix R

**Amyotrophic Lateral Sclerosis Assessment Questionnaire-5**

**(ALSAQ-5)**

**The following statements all refer to certain difficulties you may have had during the last 2 weeks. Please indicate, by ticking the appropriate box, how often the following statements have been true for you.**

*If you cannot do the activity at all*
*please tick* **Always or cannot do at all.**

## *How often during the last 2 weeks have the following been true for you?*

*Please tick* ***one box*** *for each statement.*

| | Never | Rarely | Some-times | Often | Always or cannot do at all |
|---|---|---|---|---|---|
| **1. I have found it difficult to stand up.** | ☐ | ☐ | ☐ | ☐ | ☐ |
| **2. I have had difficulty using my arms and hands.** | ☐ | ☐ | ☐ | ☐ | ☐ |
| **3. I have had difficulty eating solid food.** | ☐ | ☐ | ☐ | ☐ | ☐ |
| **4. I have felt that my speech has not been easy to understand.** | ☐ | ☐ | ☐ | ☐ | ☐ |
| **5. I have felt hopeless about the future.** | ☐ | ☐ | ☐ | ☐ | ☐ |

*Please make sure you have ticked* ***one box for each statement.***

***Thank you for completing this questionnaire.***

ALSAQ-5 Version 1.00 © 2000 Isis Innovation Ltd

# Appendix S

**ALS Functional Rating Scale**

**(ALSFRS-R)**

# ALS Functional Rating Scale

**ID No #**_____
**FC ID No** _____
**Date:**_____

The ALS Functional Rating Scale is used to assess changes in physical functioning in persons with ALS/MND. This scale is widely used by doctors and health care practitioners in clinical research and patient care in the ALS/MND community.

The following questions refer to how the person with ALS/MND is currently functioning at home. Please read each item carefully and base your answers on functioning today compared to the time before there were symptoms of ALS/MND. Please choose the answer that best fits the person with ALS/MND's functional status today.

| Domain | Level of Functional Impairment | Score |
|---|---|---|
| **Have you noticed changes in speech?** | 4- No change<br>3- Noticeable speech difference<br>2- Speech has changed; asked often to repeat words and phrases<br>1- Speech has changed; sometimes needs to use alternative communication methods (computer, writing pad, letter board, etc.)<br>0- Unable to communicate verbally | |
| **Have you noticed changes in salivation?** | 4- No change<br>3- Slight but definite excess of saliva in mouth, with or without nighttime drooling<br>2- Moderate amounts of excessive saliva, with or without minimal daytime drooling<br>1- Marked excess of saliva with some daytime drooling<br>0- Marked drooling; requires constant tissue or handkerchief | |
| **Have there been any changes in ability to swallow?** | 4- No changes (all food and liquids)<br>3- Some changes in swallowing or occasional choking episodes (including coughing during swallowing).<br>2- Unable to eat all consistencies of food and have modified the consistency of foods eaten<br>1- Use a feeding tube (PEG) to suppliement what is eaten by mouth<br>0- Eat nothing by mouth and receive all nutrition through a feeding tube (PEG) | |
| **Has there been a change in handwriting?**<br>Please choose the answer that best describes hand-writing with dominant (usual) hand without a cuff or brace. | 4- No changes<br>3- Slower and/or sloppier but all words are legible<br>2- Not all words are legible<br>1- Able to hold a pen but unable to write<br>0- Unable to hold a pen | |
| If most of meals are eaten by mouth, answer this question:<br>**Cutting Food and handling utensils**<br><br>If most of nutrition is through a feeding tube (PEG), answer this question:<br>**Use a feeding tube (PEG)** | 4- No change<br>3- Somewhat slow and clumsy, but no help needed<br>2- Sometimes need help with cutting more difficult foods<br>1- Food must be cut by someone else, but can feed without assistance<br>0- Needs to be fed<br><br>**OR**<br><br>4- Use PEG without assistance or difficulty<br>3- Use PEG without assistance, but may be slow and/or clumsy<br>2- Require assistance with closures and fasteners<br>1- Can provide minimal assistance to caregiver<br>0- Unable to perform any of the manipulations | |

| | | |
|---|---|---|
| **Has there been a change in ability to dress and perform self-care activities?** (bathing, teeth brushing, shaving, combing hair, etc.) | 4- No change<br>3- Perform self-care activities without assistance but with increased effort or decreased efficiency<br>2- Needs some help or using different methods (i.e. sitting down to get dressed, fastening buttons with non-dominant hand, etc.)<br>1- Require daily assistance<br>0- Helpless in bed | |
| **Has there been a change in ability to turn in bed and adjust the bed clothes?** | 4- No change<br>3- Can turn in bed and adjust bed clothes without assistance, but it is slower and more clumsy<br>2- Can turn in bed or adjust bedclothes without assistance but with great difficulty<br>1- Can initiate turning in bed or adjusting the bed clothes but require assistance to complete the task<br>0- Helpless in bed | |
| **Has there been a change in ability to walk?** | 4- No change<br>3- Walking has changed, but do not require any assistance or devices (foot brace, cane, walker)<br>2- Walks with assistance (cane, walker, brace, or hand held assistance)<br>1- Can move legs or stand up, but unable to walk from room to room<br>0- Cannot walk or move legs | |
| **Has there been a change in ability to climb stairs?** | 4- No change<br>3- Slower<br>2- Unsteady and/or more fatigued<br>1- Requires assistance (use of a cane, person, or handrail)<br>0- Cannot climb stairs | |
| **Is there shortness of breath or difficulty breathing?** | 4- No change<br>3- Shortness of breath only when walking<br>2- Shortness of breath with minimal exertion: talking, eating, dressing,<br>1- Shortness of breath while either sitting or lying down<br>0- Significant shortness of breath (all of the time) and considering using mechanical ventilation | |
| **Is there shortness of breath or difficulty breathing while lying down on back?** | 4- No change<br>3- Some shortness of breath while lying on back, but don't routinely use more than two (2) pillows to sleep<br>2- Some shortness of breath while lying on back, and require more than two (2) pillows to sleep<br>1- Can only sleep sitting up due to shortness of breath<br>0- Require the use of respiratory support to sleep and do not sleep without it. | |
| **Is respiratory support required?** | 4- No respiratory support<br>3- Intermittent use of BiPAP<br>2- Continuous use of BiPAP at night<br>1- Continuous use of BiPAP during night and day (nearly 24 hours)<br>0- Mechanical ventilation by intubation or tracheostomy | |
| | **Total Score** | |

**Evaluation/Comments**_____

_____

_____

_____

**Evaluation completed by:** _____

# Appendix T

**Blessed Orientation Memory Concentration Test**

**(BOMC)**

The Blessed Orientation-Memory-Concentration (BOMC) Test

"Now I'd like to give you a short memory test that will take about 5 minutes. Some questions will be easy; some may be more difficult. Are you ready?"

| Items | Maximum Error | | Score | | Weight |
|---|---|---|---|---|---|
| 1) What year is it now? | 1 | | | x4= | |
| 2) What month is it now?<br>Memory phrase:<br>Repeat phrase after me:<br>"John Brown, 42 Market Street, Chicago." | 1 | | | X3= | |
| 3) About what time is it (within 1 hour) | 1 | | | x4= | |
| 4) Count backwards 20 to 1. | 2 | | | x2= | |
| 5) Say the months in reverse order (start with December) | 2 | | | x2= | |
| 6) Repeat the memory phrase.<br>(l)John<br>(1)Brown<br>(1)42<br>(1) Market<br>(1) Chicago) | 5 | | | x2= | |
| | | | | **TOTAL** | |

The scores from each of the six items are multiplied to yield a weighted score. Score 1 for each incorrect response. Weighted error scores greater than 10 are consistent with dementia.

Scoring items 4 and 5: For uncorrected errors, score "2"; for self-corrected errors, score "1". For no errors, score "O"

Scoring the memory phrase: If no cue is necessary and the patient recalls both name and address, score "O". If patient cannot spontaneously recall the name and address, cue with "John Brown" one time only. If this cue is necessary, the patient automatically has 2 errors.

Score 1 point for each subsequent "unit" the participant cannot recall.

Source: Katzman R., et al. Validation of a short orientation-memory-concentration test of cognitive impairment. Am T Psychiatry 1983; 140:734-9.

# Appendix U

**Media Release**

# Media Release

**7 December 2012**
**C194 / 12**

## Curtin studies impact of life reflections for Motor Neurone Disease patients

Curtin Health Innovation Research Institute (CHIRI) PhD candidate, Brenda Bentley, is helping people diagnosed with Motor Neurone Disease (MND) to write their life stories and is researching the impact of this on their mental health.

Based within the WA Centre for Cancer and Palliative Care, the study looks at whether the process of reflecting on one's life in a recorded interview can relieve psychological distress as a person faces the end of their life.

"This type of therapy (coined 'dignity therapy') was developed in Canada by Dr Harvey Max Chochinov and was originally used with cancer patients and delivered good results," Ms Bentley said.

"So far, 24 people with MND have taken part in the study and in a few cases, interviews have taken place via email as some people with MND can lose the ability to speak.

"I'm hoping to encourage another 10 people to take part in the study before analysing the results and bringing the study to a conclusion early next year."

According to Ms Bentley, dignity therapy could be particularly effective for not only MND sufferers, but also their carers.

"MND is a family disease, and family carers carry an exceptional burden by providing extraordinary amounts of care. Previous research in Australia indicates that family carers provide the majority of daily care for people with MND throughout the course of the illness," she added.

"Importantly, a caregiver's distress impacts the mental and physical status of the person with MND. A palliative care intervention that assists the family carer in increasing hope may help to reduce depression and minimise distress for both parties."

Ms Bentley is hopeful the feasibility study will pave the way for future, larger studies of dignity therapy with MND populations.

The study has funding support from the Australian Research Council and the Motor Neurone Disease Association of WA.

To find out more about the study, or to take part, please contact Brenda Bentley, WA Centre for Cancer and Palliative Care, via Brenda.Bentley@curtin.edu.au.

**Contact:**

**Brenda Bentley, PhD Candidate, CHIRI - WA Centre for Cancer and Palliative Care, Curtin University**
Tel: 0427 737 712 Email: Brenda.Bentley@curtin.edu.au

**Susanna Wolz, Public Relations, Curtin University**
Tel: 08 9266 9085 Email: Susanna.Wolz@curtin.edu.au Mobile: 0401 103 877

# Appendix V

**Participant Information Sheet and Consent Form**

**National Recruitment –E-Health**

Australian Government
Australian Research Council

mnd Western Australia

WA CENTRE for CANCER and PALLIATIVE CARE

Curtin University
CURTIN HEALTH INNOVATION RESEARCH INSTITUTE

# DIGNITY THERAPY/MND STUDY

## INFORMATION SHEET – PARTICIPANTS


**INVESTIGATORS**

**Brenda Bentley, PhD Research Student, Curtin University**
**Prof. Samar Aoun, Principal Investigator, Director, Western Australia Centre for Cancer and Palliative Care, Curtin University**
**Dr. Moira O'Connor, Co-Supervisor, Western Australia Centre for Cancer and Palliative Care, Curtin University**
**Dr. Lauren Breen, Co-Supervisor, Department of Psychology and Speech Pathology, Curtin University**
**Dr. Harvey Chochinov, Associate Supervisor, Director, Manitoba Palliative Care Research Unit**


You are being asked to participate in a research study. Please take your time to review this Information Sheet and Consent Form and discuss any questions you may have with the Research Student, Brenda Bentley, mobile 0427 737 712, or email brenda.bentley@curtin.edu.au, or with the Principal Investigator of the study, Professor Samar Aoun, mobile 0419 911 940. You may take your time to make your decision about participating in this research study and you may discuss it with your regular doctor, friends and family. This consent form may contain words that you do not understand. Please ask for assistance if there is any information that you do not clearly understand.

## PURPOSE OF STUDY

The purpose of the study is to examine whether Dignity Therapy enhances the quality of life for persons and their families who are living with Motor Neurone Disease. In previous studies, Dignity Therapy has proven to be beneficial to people who have terminal illness, as well as to their family members, but most people in prior studies had cancer diagnosis. In this study, we are researching whether similar positive results will be seen with persons with Motor Neurone Disease.

A total of 50 participants diagnosed with MND from Australia will participate in this study. A family carer of each person with MND will also be invited to participate.

## STUDY PROCEDURES

If you agree to take part in this research, you will receive Dignity Therapy. In Dignity Therapy, you will have the opportunity to discuss things that you consider most important or feel are/have been most meaningful to you in life. You will be guided through a series of questions about issues that are most meaningful to you, to develop a 'life manuscript'. This includes a review of your life and your most important achievements. The therapy and all study procedures will take place either 1) through in-person meetings in your home or care setting or 2) if you are located more than 100 km outside the Perth Metropolitan area and/or have the lost the ability to speak, by utilizing the telephone, computer and/or post. All meetings will be arranged at times that are the most convenient for you.

At the beginning of the study, you will be asked to complete a series of questionnaires. It is estimated that it will take approximately 45 minutes to do this. The Research Student, Brenda

Bentley, a therapist trained in Dignity Therapy and doctoral student at Curtin University, will be available to assist you with this task either in person or by phone or videoconference.

In-Person and Video-Conferencing

If the therapy is to be conducted by in-person meetings or videoconferencing, the second meeting will be scheduled as soon as you are able to do so, ideally no more than 24 – 48 hours after your initial session with the Research Student. This session will be digitally recorded so that you can be provided with an edited transcript of this meeting for you to keep. The recorder may be shut off at any time during the course of the session, and you will have control on guiding the content of these sessions. The length of the session will be entirely determined by your energy level, engagement in the process, and wish to proceed. The Research Student will provide you only with as much guidance or assistance as you deem necessary. This taped session will likely last about 45 minutes.

The third meeting will consist of your receiving the typed, edited transcript of your therapy session from the previous meeting. You may read the transcript yourself, or if you prefer, the Research Student will read it aloud to you. This meeting will also provide you an opportunity to discuss your wishes regarding any editorial changes (additions, deletions or any clarifications) you wish to take place. This session is estimated to take 30 to 45 minutes.

At the fourth meeting, you will be provided with the final, edited Dignity Therapy transcript. This meeting is very brief.

Therapy through E-Mail

Especially if your speech is impaired, you may complete the therapy via email. If this method is chosen, you will correspond through email with the Research Student who will ask you questions and compile your answers. The Research Student will provide you with as much guidance and assistance as you deem necessary. This process of correspondence will progress according to your energy level, engagement in the process and wish to proceed, and ideally take no more than a week. The Research Student will provide you with a complete edited draft of the transcript resulting from the correspondence and offer you the opportunity for any editorial changes (additions, deletions or any clarifications) you wish to take place. You will then be provided with the final, edited Dignity Therapy transcript via post.

No matter how the therapy is delivered, one week after you receive the final document you will be provided with the opportunity to offer feedback regarding your experience and satisfaction with the Dignity Therapy approach by completing a feedback questionnaire. We will also ask you to complete the same questionnaires you completed prior to the therapy. These forms will be delivered by post or email, and will take about 45 minutes to complete.

**Family Carer Involvement**

It is important for this study that we also have contact with a family member or significant person involved with your care to gain information regarding the meaning/impact of this study for them. The Research Student will meet with your family carer where possible and ask them to complete questionnaires at the time you are entered into the study. The questionnaires may also be completed and returned via post or email. One week after completing Dignity Therapy we will also ask your family carer to complete a feedback questionnaire and the same questionnaires he/she completed prior to the therapy. These forms will be delivered by post or email, and it will take about 20 minutes to complete.

## RISKS AND DISCOMFORTS/BENEFITS

While it is possible that you may benefit from your participation in this study, it is not necessarily expected that you will; however, the study should contribute to a better understanding of support needed by persons with MND such as yourself. Participants and/or family carers may experience a stirring of emotions and feelings that you may wish to discuss further with a counsellor or other support person. The Research Student will be available to discuss this with you. In the event emotional distress occurs, you will be referred by the Research Student to the Emotional Support Program available through the Motor Neurone Disease Association of WA (MNDAWA) and the Special Care Adviser appointed to oversee this program. The Emotional Support Program is an ongoing program available to you through MNDAWA and you are able to take advantage of these services at any time, regardless of your participation in the study.

## COSTS/PAYMENT FOR PARTICIPATION

There will be no cost to you for participating in this study. You will not be paid for your participation in this study.

## CONFIDENTIALITY

All information collected by us will be treated as confidential in accordance with the National Health and Medical Research Council guidelines (NHMRC). This means that all information about you including the questionnaires will be recorded with a code number and not your name, and the consent form will be kept separate from all the information collected for the study. De-identified information gained from participants will be secured in a locked filing cabinet and stored for five years. Following this time period the information will be destroyed, all written material with be shredded, computer files will be erased. An exception to this is audio recordings, which will be erased at the conclusion of the study before being disposed of to protect identity in accordance with NHMRC guidelines. If the results of the study are published, you and your family member's identity will remain confidential.

An exception to confidentiality is if you or your family member disclose a serious intent to harm yourself or someone else, and then we have an obligation to disclose only that information to your health care team.

## VOLUNTARY PARTICIPATION/WITHDRAWAL FROM THE STUDY

Your decision to take part in this study is voluntary. You may refuse to participate or you may withdraw from the study at any time, without any consequence, jeopardy or prejudice to your future medical treatment.

## QUESTIONS

You are free to ask any questions that you may have about your rights as a research participant. If any questions come up during or after the study, please contact the **Research Student, Brenda Bentley, mobile 0427 737 712, or email brenda.bentley@curtin.edu.au, or the Principal Investigator: Professor Samar Aoun, mobile 0419 911 940.**

*This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number HR **19/2011**). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/o Office of Research and Development, Curtin University, GPO U1987, Perth, 6845 or by telephoning 9266 2784 or by emailing hrec@curtin.edu.au.*

Australian Government
Australian Research Council

mnd Western Australia

WA CENTRE for CANCER and PALLIATIVE CARE

Curtin University
CURTIN HEALTH INNOVATION RESEARCH INSTITUTE

## DIGNITY THERAPY/MND STUDY

## PARTICIPANT CONSENT FORM

Please do not sign this consent form unless you have a chance to ask questions and have received satisfactory answers to all of your questions.

1. I have read and understood the Information Sheet-Participants and Participant Consent Form, and I freely and voluntarily agree to take part in the research study called "Dignity Therapy/MND Study."
2. I understand that I will be given a copy of the signed and dated Information Sheet-Participants and Participant Consent Form. I have received an explanation of the purpose and duration of the study and the potential risks and benefits that I might expect. I was given sufficient time and opportunity to ask questions and to reflect back on my understanding of the study to the Research Student. My questions were answered to my satisfaction.
3. I am free to withdraw from the study at any time, for any reason, and without prejudice to my future medical treatment.
4. By signing and dating this document, I am aware that none of my legal rights are being waived.
5. I understand that all people participating in this study will be asked to fill out a series of questionnaires at the beginning and the end of the study.
6. I understand that I will be referred to the services of the Emotional Support Program through the Motor Neurone Disease Association of WA if I experience any distress as a result of my participation, which are available to me on an ongoing basis regardless of my participation.
7. I understand and give permission for the Research Student to contact my family carer in order to obtain feedback from them on the meaning/impact that this intervention has had on them.
8. Unless Dignity Therapy is performed via written correspondence, I understand that the Dignity Therapy interview that I will be participating in as part of this study will be audio recorded.
9. I agree to take part in this research study and for the data obtained to be published, provided my name or other identifying information is not used.

Signature of Participant: ---------- Date: ----------

Printed name of above: ----------

I confirm that I have explained the purpose and duration of this study, as well as any potential risks and benefits, to the participant whose name and signature appear above. I confirm that I believe that the participant has understood and has knowingly given their consent to participate by his/her personally dated signature.

Signature of Researcher: ---------- Date:----------

Printed name of above: ----------

# Appendix W

**Contact Sheet**

# DT/MND STUDY CONTACT SHEET

| **PARTICIPANT NO.** | **DATE:** | |
|---|---|---|
| Who was present: | | |
| Type of residence: | | |
| City: | Time Arrived: | Time Departed: |
| Initial DT1 DT2 DT3 DT4 Final Post-testing | If Final, Number of DT Transcripts _____<br>Length _____ words _____ pages | |

**OBJECTIVE EXPERIENCE**

**SUBJECTIVE EXPERIENCE**

# Appendix X

**Protocol for minimizing risk of emotional and psychological harm to participants**

**Protocol for minimizing risk of emotional and psychological harm to participants**

The methods and protocols for engaging participants in this study have been developed to minimize the risk of stress, burden and emotional harm to all participants. It is not anticipated that participants will experience emotional or psychological harm, and this has not been a factor in prior Dignity Therapy studies. While confronting the end of life may cause considerable distress in some people, the types of themes discussed in Dignity Therapy - including reflecting on memorable parts of one's life, important roles that have been played, accomplishments, things about themselves they wish to have remembered, hopes and dreams for their loved ones, words of advice and guidance - are positive and affirming themes that generally do not cause considerable distress. The research student has discussed the potential for emotional upset, as well as past experiences of this kind, with the intervention's creator, Dr. Chochinov, and with other therapists and past researchers at the Dignity Therapy training workshop, as well as with the research staff involved in the Dignity Therapy studies at the WA Centre for Cancer and Palliative Care. As a result of these investigations, the research student believes there is a very small chance of encountering significant emotional distress during this study, and that upset that may occur will be rare and minor in nature.

Nonetheless, if a participant in this study does experience distress, a protocol has been developed to ensure the participant's safety, as well as access to support services.

Creating a safe and secure environment. While engaging in Dignity Therapy, the research student/therapist will establish a trusting relationship with the participant and maintain a therapeutic presence. This includes creating trust, establishing rapport, maintaining a calm demeanour, creating safe space, establishing an empathic relationship, maintaining eye contact and engaged body language, listening deeply, and creating a safe container. To the extent possible, the intervention will take place in a private area with a minimum of noise and distraction, with the availability of a support person, such as a family member. The research student/therapist will explain to the participant before beginning Dignity Therapy that they may experience some distress at various times, and if that occurs, the research student is unable to provide therapeutic support outside of Dignity Therapy; however, support is available to them through the Emotional Support Program of the Motor Neurone Disease Association.

Upset during intervention. If a participant begins to become upset or distressed during the intervention, the research student/therapist will slow her own breathing, model calmness, observe the distress and invite the participant to discuss the memories, feelings or issues that are arising and causing the distress. If this process does not alleviate the distress and lead back to the Dignity Therapy intervention, the research student/therapist will inquire into the participant's emotional state, offer to take a break, offer to seek out the participant's support person and/or terminate the session, if appropriate, and the Action Plan will be followed.

Debriefing. At the end of the intervention, after recording has concluded and before the research student/therapist leaves the premises, the research student will inquire into how the participant is feeling. If the participant is distressed or emotionally upset, the Action Plan will be followed. The participant's support person or a family member will be sought out or contacted prior to the research student leaving the premises.

Action Plan: (Actions to be taken in all instances where emotional distress is encountered or observed).

1. A discussion will take place reiterating that the research student is unable to provide therapeutic support outside of Dignity Therapy.
2. The research student will remind the participant and family carer about the Emotional Support Program available through the Motor Neurone Disease Association of WA (MNDAWA). This program has a Special Care Adviser, Diana Menzies, responsible for counselling referrals and follow-up.
3. The research student will ask the participant if they would like me to give them the contact information for Diana Menzies, the Special Care Advisor at MNDAWA who is responsible for the Emotional Support Program, or if they would like for me to contact Diana Menzies, Special Care Adviser, on their behalf, in order to inform Diana of the participant's potential need for support.
4. The participant will also be reminded that they can withdraw from the study if the process is upsetting to them.
5. The research student will make a note to follow up with the participant by phone the following day. If the participant reports they continue to feel upset and the Special Care Adviser has not been contacted, the Action Plan will again be commenced.
6. Regular follow-up contact will also occur between appointments when the research student confirms the next appointment and also checks on the participant's physical and emotional health.
7. Follow-up contact will continue until the participant reports they are no longer experiencing distress, it has been confirmed that they are in contact with Diana Menzies and the Emotional Support Program at MNDAWA or other support services, or they do not wish to have further follow-up.

Letter two weeks after intervention. Two weeks after the final Dignity Therapy visit, and one week after post-testing, the research student will send a letter thanking the participants for their participation. The letter will contain a reminder about the Emotional Support Program available through MNDAWA, including contact information.

# Appendix Y

**Permission to reproduce article: Bentley, B. "It takes the time that it takes," Journal of Palliative Medicine, 15(8), 949-950**

**Brenda Bentley**

**From:** Ballen, Karen <KBallen@liebertpub.com>
**Sent:** Monday, 16 June 2014 10:26 PM
**To:** Brenda Bentley
**Subject:** FW: Journal of Palliative Medicine - Decision on Manuscript ID JPM-2012-0054
**Attachments:** winmail.dat; ATT00001..htm

Dear Brenda:

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Kind regards,

Karen Ballen

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-----Original Message-----
From: Brenda Bentley [mailto:brenda.bentley@postgrad.curtin.edu.au]
Sent: Sunday, June 15, 2014 8:52 PM
To: Bicovny, Esther
Subject: RE: Journal of Palliative Medicine - Decision on Manuscript ID JPM-2012-0054

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Thank you.

Kind regards,
Brenda

# Appendix Z

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## Brenda Bentley

**From:** BMCSeriesEditorial <BMCSeriesEditorial@biomedcentral.com>
**Sent:** Tuesday, 29 July 2014 5:37 AM
**To:** Brenda Bentley
**Subject:** RE: Permission

Dear Dr. Bentley,

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Please let us know if you have any further questions.

Sincerely,
MJ
---

**Maria Merrie Jul Ladag**
Journal Editorial Office
**BioMed Central**

E: editorial@biomedcentral.com
W: www.biomedcentral.com
**********

---

**From:** BMCSeriesEditorial
**Sent:** 23 July 2014 16:31
**To:** 'Brenda Bentley'
**Subject:** RE: Permission

Hi Dr. Bentley,

Thank you for your email and letting us know about your concern. Kindly be informed that your query has been forwarded to our handling Editor for advice and as soon as we hear from her, we will get in touch.

Hope this helps. Should you have further questions, please don't hesitate to contact me.

Sincerely,
MJ
---

**Maria Merrie Jul Ladag**
Journal Editorial Office
**BioMed Central**

E: editorial@biomedcentral.com
W: www.biomedcentral.com
**********

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**Brenda Bentley**

**From:** noreply@salesforce.com on behalf of Karen Baulch <plosone@plos.org>
**Sent:** Monday, 28 July 2014 2:12 AM
**To:** Brenda Bentley
**Subject:** Permission ref:_00DU0lfis._500U0DNl1C:ref

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# Appendix AA

**Correlation Tables**

**Table AA-1: Means, Standard Deviations, and Intercorrelations of Outcome Measures for People with MND**

| Measure | N | M | SD | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 |
|---|---|---|---|---|---|---|---|---|---|---|---|
| **1. ALSAQ 1** | 28 | 9.32 | 4.04 | | | | | | | | |
| **2. FACIT-sp 1** | 29 | 30.72 | 10.43 | -.20 | | | | | | | |
| **3. HHI 1** | 29 | 38.76 | 5.10 | -.21 | .77** | | | | | | |
| **4. PDI 1** | 29 | 48.59 | 15.45 | .55** | -.51** | -.41* | | | | | |
| **5. ALSAQ 2** | 26 | 9.35 | 3.71 | .80** | -.32 | -.37 | .49* | | | | |
| **6. FACIT-sp 2** | 27 | 30.92 | 9.88 | -.24 | .83** | .59** | .44* | -.35 | | | |
| **7. HHI 2** | 27 | 36.61 | 6.80 | -.30 | .36 | .37 | -.27 | -.28 | .43* | | |
| **8. PDI 2** | 24 | 47.59 | 12.91 | .47* | -.36 | -.30 | .87** | .49* | .36 | -.15 | --- |

** Correlation is significant at the 0.01 level (2-tailed).

* Correlation is significant at the 0.05 level (2-tailed).

**Table AA-2: Means, Standard Deviations, and Intercorrelations of Outcome Measures for MND Family Carers**

| Measure | N | M | SD | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| **1. ALSFRS-R 1** | 18 | 32.61 | 9.76 | | | | | | | | | | |
| **2. ZBI 1** | 18 | 12.44 | 7.89 | -.40 | | | | | | | | | |
| **3. HHI 1** | 18 | 38.39 | 4.46 | .03 | -.38 | | | | | | | | |
| **4. HADS-A 1** | 18 | 7.28 | 3.71 | -.30 | .83** | -.58* | | | | | | | |
| **5. HADS-D 1** | 18 | 4.17 | 3.33 | -.60** | .72** | -.40 | .66** | | | | | | |
| **6. ALSFRS-R 2** | 17 | 30.12 | 9.62 | .95** | -.38 | -.03 | -.22 | -.50* | | | | | |
| **7. ZBI 2** | 17 | 16.29 | 11.22 | -.39 | .82** | -.31 | .76** | .58* | -.482 | | | | |
| **8. HHI 2** | 17 | 36.71 | 4.52 | -.04 | -.20 | .62** | -.31 | -.09 | -.014 | -.25 | | | |
| **9. HADS-A 2** | 17 | 6.88 | 4.33 | -.41 | .82** | -.25 | .86** | .62** | -.420 | .78** | -.15 | | |
| **10. HADS-D 2** | 17 | 4.41 | 3.91 | -.60* | .79** | -.25 | .70** | .80** | -.615** | .86** | -.22 | .74** | ---- |

** Correlation is significant at the 0.01 level (2-tailed).

* Correlation is significant at the 0.05 level (2-tailed).